

**THE PROCESS OF ADHERING TO AEROSOL THERAPY IN
ADOLESCENTS WITH CYSTIC FIBROSIS: PATIENT AND PARENT
PERSPECTIVES**

Daniel Patrick Harry O'Toole

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ABSTRACT

Advances in medicine and healthcare have resulted in significant improvements for those children and adolescents living with a chronic health condition. However, the treatments prescribed will only work if they are taken. Rates of adherence to treatment in cystic fibrosis are low. This is especially true for adherence to aerosol therapy, a particular concern considering the reliance on aerosol therapy for the treatment of *Pseudomonas aeruginosa*.

The advent of Adaptive Aerosol Delivery™ (AAD) technology has enabled accurate and objective measurement of adherence to aerosol therapy treatment. However, the factors which influence the process of adhering to aerosol therapy remain largely elusive. Therefore, the current study aims to qualitatively explore the process of adhering to aerosol therapy in a clinical sample of adolescents with CF from both a patient and parent perspective. In addition, objective data downloaded from an AAD device (the I-neb™, Respironics, Chichester, UK) will be used to cue reflections on actual events around adherence.

12 participants were interviewed (six parent-child dyads). The participating adolescents were all aged between 11 and 16 years and had a CF diagnosis. There were five mothers and one father who took part. The parent and adolescent data were analysed separately using the Grounded Theory Method. Nine core categories for the parent data and nine core categories for the adolescent data were developed into a coherent framework and represented as a theoretical formulation which described the process of adhering to aerosol therapy from both a parent and adolescent patient perspective. The theoretical formulation highlights the complexity involved and details the numerous interacting biological, psychological, social and environmental influences on adherence to aerosol therapy. These findings are discussed in relation to the existing literature and clinical implications are considered. The results of the current study complement and extend previous research on adherence in CF.

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1.0 INTRODUCTION

1.1 Children and adolescents with a chronic health condition

Estimates of the numbers of children and adolescents living with a chronic health condition are not easy to establish. The wide variety of data collection methods being utilised (e.g., medical files, questionnaires, lists of hospital diagnostic codes) and the considerable discrepancies regarding the definition of what constitutes a chronic health condition, makes establishing prevalence rates difficult (Zylke & DeAngelis, 2007). The terms “chronic disease” and “chronic illness”, for example are often used interchangeably. Williams (1997) defines a chronic disease as a “medically diagnosed ailment with a duration of six months or longer which shows little change or slow progression” (pg. 312). Pless and Douglas (1971) define a chronic illness as a “physical, usually non-fatal condition that has lasted longer than three months in a given year or necessitated a period of continuous hospitalisation of more than one month; of sufficient severity to interfere with the child’s ordinary activities to some degree.” The difficulties associated with measuring and defining chronic health conditions in children and adolescents led to a recent systematic review by van der Lee and colleagues (van der Lee, Mokkink, Grootenhuys, Heymans & Offringa, 2007) finding prevalence rates ranging from as little as 0.22% to upwards of 44%. Although they were unable to establish a firm estimate, they suggest that prevalence rates of children and adolescents living with a chronic health condition have increased since the 1980’s and predict likely increases in the future.

Spinks (2009) provides a useful list of examples of chronic health conditions which include: cystic fibrosis (CF), epilepsy, asthma, diabetes, sickle cell disease, leukaemia and other childhood cancers. The disease burden placed upon individual children living with these conditions and their families is substantial (Taylor, 2006). In addition, the increases in prevalence rates bring significant organisational and financial consequences, particularly for educational provision, health care planning and employment (Newacheck, & Halfon, 1998; van der Lee et al., 2007).

1.2 Psychosocial aspects of chronic health conditions

The majority of children, adolescents and their families cope well with the demands of living with a chronic health condition (Taylor, 2006; Spinks, 2009). Although that may be the

case, chronic health conditions can contribute to a number of psychosocial difficulties. Barlow & Ellard (2006), for example, suggest that fatigue, pain and other physical symptoms, combined with the need for disease management regimes, may well impede many aspects of daily living for children, parents and siblings. Children and adolescents face a number of specific challenges including having to adhere to an often complex treatment regimen, coming to terms with parental concerns, and managing worries that they may have regarding their own self-image (Miller & Wood, 1991). Children and adolescents with a chronic health condition can spend a significant amount of time in the hospital setting. Pao and colleagues (Pao, Ballard, & Rosenstein, 2007) investigated the impact of what they described as “growing up in the hospital” and suggested that the effects on emotional, cognitive, and social development can be profound.

Many of the above challenges place children and adolescents living with a chronic health condition at greater risk for developing emotional and behavioural adjustment problems. There is evidence of a two-fold increase in risk of psychological maladjustment when compared to healthy controls (Lavigne & Faier-Routman, 1992; Hysing, Elgen, Gillberg, Lie, & Lundervold, 2007). In addition, it has been suggested that children and adolescents living with a chronic health condition are placed at greater risk of experiencing an overall lowered quality of life (Lebovidge, Lavigne, Donenberg, & Miller, 2003; Garnefski, Koopman, Kraaij, & ten Cate, 2009).

The impact on families of caring for a child or adolescent with a chronic health condition should not be understated. Coping with the initial diagnosis can come as a major shock (Cherry, 1989) and parents can experience anxiety about their child’s future prognosis and overall well-being (Barlow & Ellard, 2006). Caring responsibilities can place heavy practical and emotion demands on parents (Magrab, 1985; Eiser, 1990). Complex treatment regimens and specific requirements associated with chronic health conditions such as CF and diabetes can result in daily care needs becoming more demanding, particularly as the condition progresses or worsens (Boekaerts & Röder, 1999). Balancing the above healthcare needs with daily work commitments and the needs of other members of the family can create an additional burden. Siblings may receive less time and attention from parents and family days out can be disrupted or made impossible (Foster, Eiser, Oades, Sheldon, Tripp, Goldman, Rice & Trott, 2001; Barlow & Ellard, 2006).

In taking the above into account, it is perhaps unsurprising that caring for a child with a chronic health condition has been associated with higher rates of marital stress, divorce, paternal unemployment and financial difficulties (Reichman, Corman, & Noonan, 2004; van Dyck, Kogan, McPherson, Weissman, & Newacheck, 2004; Noonan, Reichman, Corman, 2005).

1.3 Adherence to treatment in chronic health conditions

The World Health Organisation (WHO) has defined adherence as “the extent to which a person’s behaviour – taking medication, following a diet, and/or executing lifestyle changes – corresponds with agreed recommendations from a health care provider” (WHO, 2003 pg. 3). Poor adherence to treatment in chronic health conditions is a significant problem with adherence rates of 30% to 70% often being cited (Meichenbaum & Turk, 1987; Rapoff, 1999; Horne, 2007). Sub-optimal adherence is costly from a healthcare perspective and can contribute to a number of serious consequences from a patient perspective including increased morbidity, mortality and lowered quality of life (DiMatteo, Giordani, Lepper & Croghan, 2002; WHO, 2003).

1.3.1 Correlates of adherence to treatment

By gaining a better understanding of the factors impacting adherence to treatment in chronic health conditions, interventions can be designed to improve it. In turn, this should reduce the disease-related mortality, morbidity and unnecessary health-care costs discussed above. In contrast to the adult literature, adherence to treatment for child and adolescent chronic health conditions has rarely been examined in relation to extant theoretical models. Therefore, the following section will begin by discussing the research on correlates of adherence to treatment for children and adolescents before moving on to briefly describe some of the existing theoretical models that are primarily used in the examination of adherence to treatment for adult populations. The research examining the interventions designed to increase adherence to treatment will also be discussed.

1.3.1.1 Patient and family correlates

Despite decades of research investigating the types of patients that are most likely to be non-adherent, little evidence has been found in relation to specific personality characteristics having an influence on adherence behaviour (McKim, Stones & Kozma, 1990; Bosley, Fosbury

& Cochrane, 1995). However, age has consistently been shown to have an impact on adherence. It is more likely, for example, that rates of adherence will be lower in adolescents than in younger children (Jacobson, Hauser, Wolfsdorf, Houlihan, Milley, & Hrskowitz, 1987; Brownbridge & Fielding, 1994; Walders, Kopel, Koins-Mitchell, & McQuaid, 2005; Feldman, De Civita, Dobkin, Malleson, Meshefedian, & Duffy, 2007). The WHO defines adolescence as the period between 10 and 19 years of age (WHO, 2008). It is a particular concern that rates of adherence are poor during this time because adolescence is the developmental stage associated with taking a greater responsibility for one's own health in preparation for adulthood (Sarafino, 2004; Berk, 2006). Studies examining patient gender as a correlate of adherence are equivocal. Although there are studies that found males to be less adherent than females to CF and diabetes regimens (Lorenz, Christensen, & Pichert, 1985; Patterson, 1985; Naar-King, Idalski, Ellis, Frey, Templin, & Cunningham, 2006) an equal number of studies found that females were less adherent than males (Johnson, Freund, Silverstein, Hansen, & Malone, 1990; Patino, Sanchez, Eidson, & Delamater, 2005; Stewart, Lee, Waller, Hughes, Low, & Kennard, 2003).

It has been consistently shown that patients with less knowledge about their treatment are less adherent (Gudas, Koocher, & Wypij, 1991; Holmes, Chen, Streisand, Marschall, Souter, & Swift, 2006; Tebbi, Cumings, Kevon, Smith, Richards, & Mallon, 1986). There are studies however, that have shown patients with greater knowledge are not necessarily more adherent (Beck, Fennell, Yost, Robinson, Geary, & Richards, 1980; McQuaid, Kopel, Klein, & Fritz, 2003), suggesting that additional factors are implicated in the process of adherence to treatment. Indeed, some have argued that one consequence of a belief in the simplistic notion that increased knowledge about treatment leads to increased adherence is that the development of more sophisticated interventions, based on detailed theoretical models of health behaviour have been held back (Horne, 2007).

In general, lower socioeconomic status and more specifically, lower parental education levels have been associated with poor adherence to regimens for CF, diabetes, renal disease and juvenile rheumatoid arthritis (Bobrow, AvRuskin, & Siller, 1985; Brownbridge & Fielding, 1994; Denson-Lino, Willies-Jacobo, Rosas, O'Conner, & Wilson, 1993; Patterson, 1985; Radius, Marshall, Rosenstock, Drachman, Schuberth, & Teets, 1978; Rapoff, Belmont, Lindsley, & Olson, 2005). Although members from black and minority ethnic groups – particularly African-American and Hispanic – have been found to be less adherent to treatment (Snodgrass,

Vedanarayanan, Parker, & Parks, 2001; McQuaid et al., 2003), some argue that the use of *race* as a predictor of non-adherence is too simplistic. Tucker et al (Tucker, Fennell, Pedersen, Higley, Wallack, & Peterson, 2002), for example, suggest that a “culturally sensitive model” should be applied, where factors related to adherence *within* different racial groups and not *between* them are studied. Further studies have demonstrated that parental divorce (Brownbridge & Fielding, 1994; Shemesh, Shneider, Savitzky, Arnott, Gondolesi, & Krieger, 2004) and patients in larger families or where mothers work outside of the home (Patterson, 1985; Tebbi et al., 1986) are less likely to be adherent to treatment regimens for cystic fibrosis and cancer.

Adherence to treatment is less likely in those patients with emotional or behavioural difficulties (Brownbridge & Fielding, 1994; Greening, Stoppelbein, Konishi, Jordan, & Moll, 2007). This finding has been examined in greater detail (through the use of meta-analytic techniques) by DiMatteo and colleagues (DiMatteo, Lepper & Croghan, 2000) who found that depression is associated with poorer rates of adherence to a wide range of treatments. The use of *denial* has been associated with lower adherence (Tamaroff, Festa, Adesman & Walco, 1992). Greater pessimism has been related to non-adherence to CF treatment regimens (Gudas et al., 1991). In addition, simply forgetting to take medications is one of the most frequently reported barriers by patients for a number of different chronic health conditions (Tucker et al., 2001; Shemesh et al., 2004; Modi & Quittner, 2006). On the more positive side of patient adjustment and coping, higher self esteem (Friedman, Litt, King, Henson, Holtzman, & Halverson, 1986), a sense of optimism (Gudas et al., 1991), increased autonomy and independence (Friedman et al., 1986), greater self-efficacy (Holmes et al., 2006), and greater problem-solving skills (McCaul, Glasgow, & Schafer, 1987) have all been associated with better adherence to treatment.

A similar situation has been found in terms of positive family adjustment and coping. Perhaps unsurprisingly, greater family support, integration, expressiveness, harmony and organisation, have all been associated with improved rates of adherence (Friedman et al., 1986; McCaul et al., 1987). Better family communication and problem solving have also been shown to have a positive effect on adherence to treatment (Bobrow et al., 1985; McQuaid, Walders, Kopel, Fritz & Klinnert, 2005). More negative aspects of coping such as increased parental stress and poor coping, have been associated with lower adherence (Brownbridge & Fielding, 1994; Gerson, Furth, Neu, & Fivush, 2004). Studies have found the same effect for poor communication (Tubiana-Rufi, Moret, Czernichow, & Chwalow, 1998; Lewandowski & Drotar,

2007), increased parental depression (Bartlett, Krishnan, Riekert, Butz, Malveaux, & Rand, 2004) and greater parental anxiety (Hazzard, Hutchinson, & Krawiecki, 1990).

1.3.1.2 Disease-related correlates

In general, chronic health conditions of longer duration are associated with lower adherence (Brownbridge & Fielding, 1994; Rapoff, 2010). In diabetes, research indicates that adherence deteriorates significantly over time, with the emergence of non-adherence being particularly evident an average of three and half years after the onset of the disease (Jacobson, Hauser, Lavori, Wolfsdorf, Herskowitz, & Milley, 1990). There tends to be an oscillation of symptoms over time with many chronic health conditions and Rapoff (1989, pg.42) suggests that “adherence may be particularly difficult to sustain during periods when patients are relatively asymptomatic.” Intuitively, the severity and frequency of symptoms and greater disease severity overall should motivate adherence in patients as they are likely to want to improve their situation. Although greater disease severity has been related to higher adherence (Rapoff et al., 2005), it is not always the case. Some studies have shown for example, that increased symptom frequency and higher disease severity is associated with lower adherence to treatment (Brownbridge & Fielding, 1994; Feldman et al., 2007). However, it may well be likely that because the above studies assessed adherence and symptoms concurrently rather than longitudinally, lower adherence actually produced the increased symptom frequency and overall deterioration.

The *actual* disease severity is not the only factor which can have an influence on adherence to treatment. The patient and parent *perceptions* of severity have also been found to play an important role. For example, the perceived severity of disease as reported by patients and parents were found to have greater predictive validity of adherence than those of providers (Rapoff & Barnard, 1991). In addition, while patient perceptions of higher severity have been associated with lower adherence (Gudas et al., 1991) maternal perceptions of higher severity have been associated with better adherence (Radius et al., 1978). Although increased parental monitoring and encouragement have been implicated in the explanation for the above findings, the fact that some studies suggest that patient and parent perceptions are differentially related to adherence have yet to be fully explained.

1.3.1.3 Regimen-related correlates

Treatment regimens that are highly complex, long-term and interfere with other aspects of a person's life may act as a barrier and have been associated with low levels of adherence (Turk & Meichenbaum, 1991; Lemanek, Kamps, & Chung, 2001; Feldman et al., 2007; April, Feldman, Zunzunegui, & Duffy, 2008). In countries that do not provide state-run healthcare, treatment costs can be prohibitive for some families. One study, for example, found that for parents with children diagnosed with paediatric rheumatic diseases, 41% said they had difficulty paying for physician-related charges and 25% said they had problems paying for medications (McCormick, Stemmler, & Athreya, 1986). In contrast, a more recent study found that lower medication costs were related to higher adherence for children with asthma (Bender, Pedan, & Varasteh, (2006).

Many treatments for chronic health conditions produce negative side effects. This is not a phenomenon restricted only to chemically derived treatments. Chest physiotherapy for patients with CF, for example, helps to effectively clear the lungs of excessive mucus but results in paroxysms of coughing and gagging (Allen, Panitch & Rubenstein, 2010). However, surprisingly few studies have examined the influence of side-effects on adherence to treatment in children and adolescents. In those that have, unpleasant-tasting medications have been related to lower adherence (Radius et al., 1978; Celano, Geller, Phillips, & Ziman. 1998). Patient and parent perceptions of the efficacious nature of prescribed treatments have also been found to influence adherence. In asthma and diabetes for example, higher levels of perceived benefits as rated by patients and parents have been associated with improved adherence (Bobrow et al., 1985; McCaul et al., 1987).

1.3.2 Theories and models of adherence to treatment

1.3.2.1 The Health Belief Model

Social cognition models (i.e., those predominantly based on the assumption that the way in which people behave in social situations is a function of rational information processing and perceived individual cognitions) examine the precursors and predictors to health behaviours. Over the past five decades, the Health Belief Model (HBM: Rosenstock, 1974; Janz & Becker, 1984) has been one of the most widely used theories in health behaviour research. Although originally developed in the 1950s in order to predict preventative health behaviours (such as the

uptake of preventative screening programmes for hypertension), the HBM was later extended in an attempt to better understand adherence to prescribed treatment regimens (Taylor, 2006).

In addition to specific demographic variables, the HBM posits five major components that help to predict or explain adherence (see Figure 1): Firstly, “perceived susceptibility” (including the patient’s perceived risk of contracting or re-contracting a condition or the level of acceptance associated with an existing condition); secondly, “perceived severity” (which includes the patient’s evaluation of the medical and social consequences of contracting an illness or not receiving appropriate treatment); thirdly, “perceived benefits” (including the patient’s judgement of the likely benefits of adhering to medical advice or treatment); fourthly, “perceived barriers” (including the patient’s perception of obstacles weighed up against the likely benefits of adhering to medical advice or treatment); and finally, “cues to action” (internal and external cues, such as increased symptoms or being reminded by others).

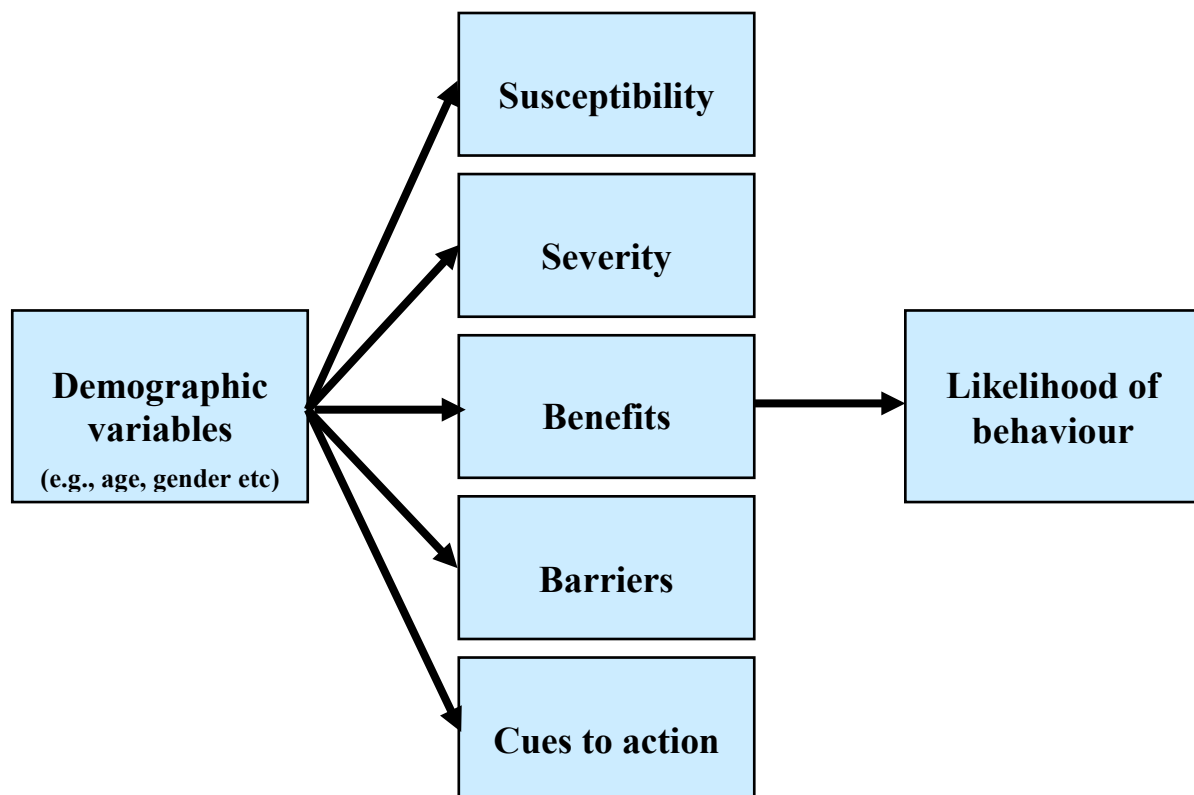


Figure 1: The Health Belief Model

There have been several studies in the adult literature that support the predictions of the HBM for a wide range of health practices (Becker, 1974; Norman & Fitter, 1989; Harrison, Mullen, & Green, 1992). There is also correlational support for components of the HBM in the child and adolescent adherence literature (Rapoff, 2010). Although the HBM is one of the only health behaviour theories to have been adapted for use within child and adolescent chronic health (The Children's Health Belief Model: CHBM), just one analogue study has tested the model's predictive validity. Bush & Iannotti (1990) found that 63% of the variance in children's expected use of medication was predicted by the CHBM, with *perceived severity* and *perceived benefit* accounting for the majority of this.

Despite the above evidence, the HBM has been criticised on a number of grounds including for: the variations in the way the model's variables have been conceptualised and measured; the focus placed on the conscious processing of information; its emphasis on the individual without due consideration for the influence of wider social and environmental influences; the way it suggests that behaviours can be predicted by attitudes and beliefs despite evidence to the contrary; and for the absence of the role of emotional factors such as fear and denial (Stroebe & Stroebe, 1995; Ogden, 2004; Rapoff, 2010).

1.3.2.2 The Theory of Planned Behaviour

The Theory of Planned Behaviour (TPB: Ajzen, 1988) is an extension of the earlier Theory of Reasoned Action (TRA, Fishbein & Ajzen, 1975) and stresses behavioural intentions as the outcome of a permutation of numerous beliefs. The theory suggests that intentions should be conceptualised as “plans of action in pursuit of behavioural goals” (Ajzen & Madden, 1986, pg. 455) and are determined by three major factors (see Figure 2): “attitude towards the behaviour” (including both positive and negative beliefs regarding the performing of a specific behaviour and the potential consequences involved); “subjective norms” (incorporating the perceived views of important people in the person's life about whether they approve or disapprove of the behaviour in question and the person's motivation to comply with these views); and “perceived behavioural control” (the individual's perception as to whether performing the behaviour is easy or difficult based on a consideration of internal skills and abilities and external obstacles and opportunities, both of which are influenced by previous behaviour).

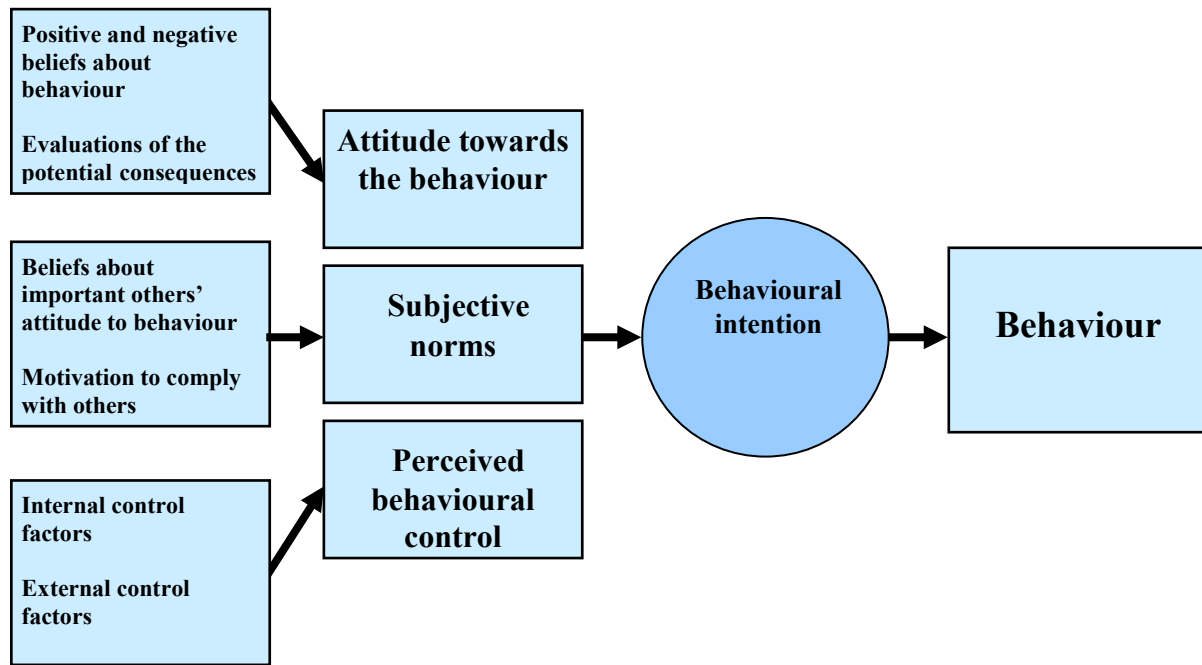


Figure 2: The Theory of Planned Behaviour

The model has been used to assess a variety of health-related behaviours and, in general, correlational studies have found support for the major components of the theory (Ajzen, 1991; Armitage & Conner, 2001). However, there have been remarkably few experimental tests of the TPB (Sutton, 2002). Studies examining the TPB in the context of adherence, particularly for children and adolescents, are also limited. Nonetheless, there are studies which have indicated that *normative beliefs* may influence adherence (Cochrane & Gitlin, 1988; Conner & Sparks, 2005). Despite its promise, the lack of experimental studies testing the model's predictive validity has been a major source of criticism. In addition, Rapoff (2010) suggests that "like all attitudinal theories, the burden is on proponents of the TPB to show that experimental manipulations designed to change attitudinal variables actually result in behaviour change" (pg.57). This is problematic when taking into account a recent meta-analysis (Webb & Sheeran, 2006) which found that a medium-to-large change in intention ($d=0.66$) led to only a small-to-medium change in behaviour ($d=0.36$). The authors concluded that although intentions do have a significant impact on behaviour, they are less important than what correlational studies have previously suggested.

1.3.2.3 The Self Regulatory Model

The Self Regulatory Model (SRM: Leventhal, 1993; Leventhal, Zimmerman, & Gutmann, 1984) is based on approaches to problem solving and attempts to explain the way in which individuals perceive, conceptualise (form representations of a health threat) and cope with the problem of illness (including their emotional response to the health threat) in order to re-establish their state of health. The model incorporates three main stages of adaptation to illness (see Figure 3): “interpretation” (the patient’s attempts to make sense of their perceived symptoms); “coping” (adaptive and maladaptive ways of dealing with the problem in order to regain a sense of balance); and “appraisal” (the assessment of how successful, or otherwise that the coping stage has been). The model is described as “self-regulatory” because the above three components interrelate in order to maintain a state of equilibrium (i.e., health). The model proposes that illness disrupts normality and the individual is motivated to return to a “normal”, healthy state.

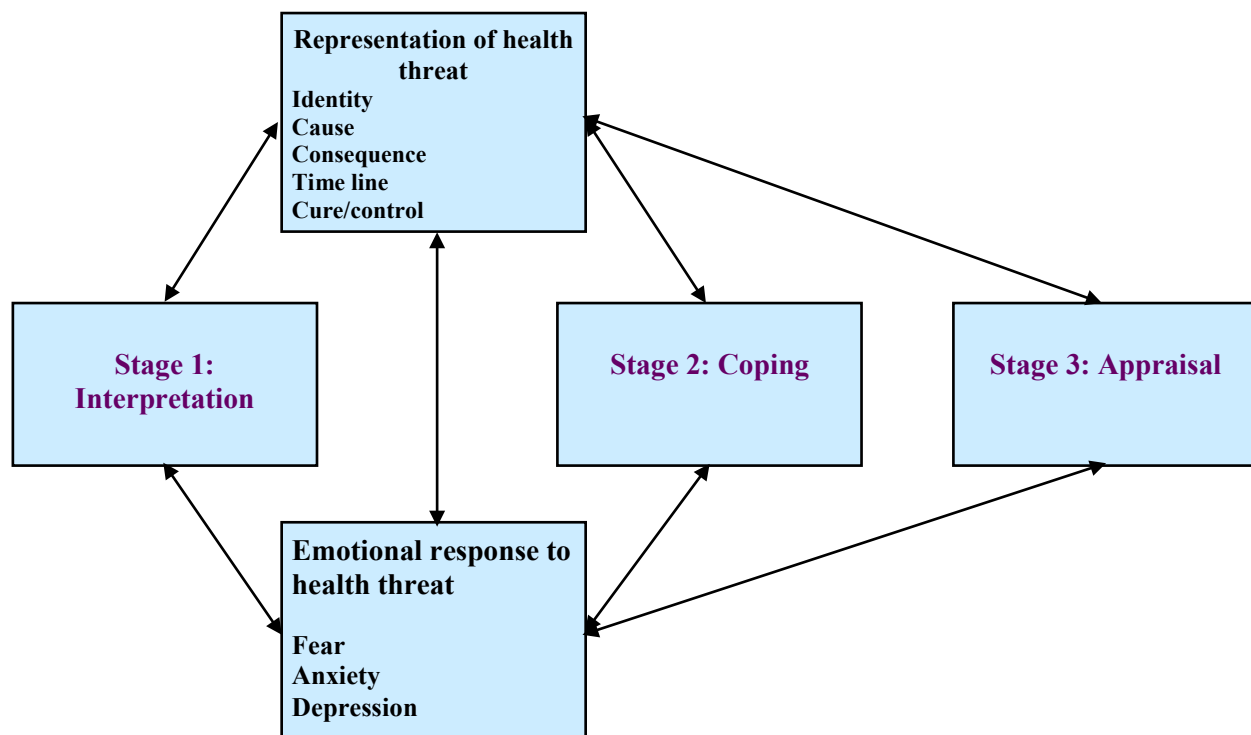


Figure 3: The Self Regulatory Model

A meta-analytic review of the SRM (Hagger & Orbell, 2003) demonstrated theoretically predictable relations between illness cognitions, coping and outcomes across the reviewed studies. In addition, the review also indicated that individual tests of illness cognitions, coping behaviours and illness outcomes found that moderate-to-strong relationships existed between these variables. In terms of adherence, Gonder-Frederick and Cox (1991) found that patients with diabetes used perceived symptoms to indicate their blood glucose levels and to guide self-treatment. A further study demonstrated that patients who believed their hypertension was an acute condition were less likely to continue taking their preventative medication than those who believed it to be a chronic condition (Meyer, Leventhal, & Guttman, 1985). However, there have been a number of studies that have failed to show direct relationships between illness perceptions and levels of adherence, leading to criticisms of the SRMs utility and forcing research attempts to focus more on specific beliefs about treatment in a search for improved cognitive predictors (Marteau & Weinman, 2004).

1.3.3 Interventions to improve adherence to treatment

Although the above research and theoretical models may have advanced our understanding about the factors which influence adherence to treatment, research efforts have also concentrated on the ways in which adherence to treatment can be improved. Much of this research has focussed on adult populations (Peterson, Takiya, Finley, 2003; Haynes, Yao, Degani, Kripalani, Garg, & McDonald 2005; Kripalani, Yao, & Haynes, 2007; Williams, Manias & Walker, 2008). However, the evidence base regarding the effectiveness of interventions designed to increase adherence in children and adolescents is beginning to grow. Although strategies that address issues such as depression or family systems difficulties have been proposed, adherence intervention strategies are usually categorised into three main groups: educational (i.e., providing more detailed information regarding treatment instructions or how to use medical equipment); organisational (i.e., improving healthcare provider communication or simplifying medical regimens); and behavioural (i.e. matching adherence behaviour with positive or negative consequences or modifying the environment to encourage better adherence) (Lemanek et al., 2001). A recent review which meta-analysed 71 studies found that adherence to interventions across all three categories (educational, organisational and behavioural) increased adherence and resulted in positive health benefits for children with chronic illnesses (Graves,

Roberts, Rapoff, & Boyer, 2010). In addition, the authors of the study found that the impact on health outcomes may be more compelling when educational and behavioural strategies were combined. They did note however, that participant and methodological characteristics appear to influence intervention effect sizes and caution of the existence of significant heterogeneity within the data sets.

Intervention studies are often hampered by difficulties in defining what constitutes adherence, the methods used to assess adherence and the designs used to assess multi-component intervention programmes (Lemanek et al., 2001). In addition, van Dulmen and colleagues (van Dulmen, Sluijs, van Dijk, de Ridder, Heerdink, & Bensing, 2007) have criticised intervention studies for failing to provide a theoretical explanation of the operating mechanisms at work, resulting in an overall lack of explanatory power. They suggest that a general lack of specific theories that adequately predict and explain non-adherence is one of the reasons for this. Overall, despite good progress having been made it has been suggested that there is still much to achieve with regards to adherence interventions for children and adolescents with a chronic health condition (Butz, 2006; Graves et al., 2010; Kahana, Drotar, & Frazier, 2008; Pai & Drotar, 2010).

1.3.4 Summary

Poor adherence to treatment in childhood chronic health conditions is a significant problem and can contribute to a number of serious consequences including increased morbidity, mortality and lowered quality of life. Correlational research has demonstrated that adherence is influenced by a number of patient and family, disease-related and regimen-related attributes. A number of theories and models have been proposed which attempt to better explain and predict the likelihood of adherence. However, research on the effectiveness of these models is predominantly focussed on the adult population with fewer studies examining their use for child and adolescent adherence. Although the models and research have advanced our understanding and enabled the development of interventions designed to improve adherence, much has yet to be achieved in this area and adherence problems prevail.

1.4 Cystic fibrosis

Cystic fibrosis (CF) is the most common life-threatening inherited disease amongst Caucasians (Davis, 2001; Walters & Mehta, 2007). The disease is characterised by generalised

dysfunction of the exocrine glands and predominantly affects the respiratory and digestive systems. With approximately 4% of the UK population carrying the recessive gene, birth prevalence rates in the UK are frequently cited as 1:2,000/2,500 live births (Bobadilla, Macek, Fine & Farrell, 2002; Dodge, Lewis, Stanton, & Wilsher, 2007). CF was once regarded as an almost universally fatal childhood genetic disorder (Allen, et al., 2010) but life expectancy for people with CF has vastly improved in recent years, attributable largely to improvements in the management and treatment of the condition. However, the benefits of improved treatment are dependent on adherence to the regimen, which remains problematic.

1.4.1 Adherence to treatment in CF

The daily treatment regimen for people with CF is burdensome and includes dietary management, vitamin supplementation, pancreatic enzyme replacement therapy, oral antibiotics, aerosol therapy (e.g., nebulised antibiotics, mucolytics and bronchodilators), exercise, chest physiotherapy and postural drainage (Arias Llorente, Bousoño Garcia, & Diaz Martin, 2008; Thomson & Harris, 2008; McColley, 2010). The situation is compounded for those individuals with additional co-morbidities (such as CF-related diabetes), as further treatments are also then required. In the CF population, adherence to the treatment regimen is generally low with estimates varying according to age, type of treatment, and method of measurement (Abbott, Dodd & Webb, 1996; Zindani, Streetman, Streetman & Nasr, 2006; Barker & Quittner, 2010). Adherence to dietary recommendations, for example, has been recorded at 40% to 50% for adults (Abbott, Dodd, Bilton, & Webb, 1994) with rates of 16% to 20% being found in children (Anthony, Paxton, Bines & Phelan, 1999). In child and adolescent populations adherence to pancreatic enzyme replacement therapy is reportedly very high (Modi, Lim, Yu, Geller, Wagner & Quittner, 2006) whereas particularly low levels are reported for chest physiotherapy (Quittner, Drotar, Ievers-Landis, Seider, Slocum, & Jacobsen, 2000). In a study of children with CF designed to examine adherence to a number of treatment elements using multiple methods of assessment, Modi and colleagues (Modi et al., 2006) reported adherence rates ranging from 67% to 100% for self-report measures and 22% to 71% for more objective measures (e.g., pharmacy refill history and electronic monitors). Each of the methods of measurement described in the above study has their own relative strengths and weaknesses and have been utilised in numerous studies investigating adherence in the CF population. The variability of the methods of

measurement used may be an important factor when accounting for the inconsistency observed in studies of adherence (Modi et al., 2006; Duff & Latchford, 2010a).

1.4.1.1 Measuring adherence to treatment in CF

Patient and parent self-report is practical and inexpensive and is the most commonly used method of measuring adherence (Bryon, 1998; Rapoff, 1999; Burkhart & Dunbar-Jacob, 2002). Although the method is flexible and can be adapted to measure all of the treatment regimen components, self-report is often unreliable. The measure is subjective and has been shown to be highly influenced by social desirability and recall biases leading to an over-estimation of reported adherence (Quittner, Modi, Lenmanek, Ievers-Landis, & Rapoff, 2008; Barker & Quittner, 2010).

The daily phone diary (DPD) method has been utilised extensively in US CF populations and attempts to elicit information regarding activities over the last 24 hours in an less obtrusive manner (Quittner et al., 2008). In doing this, the DPD method attempts to reduce social desirability bias and has been reported as demonstrating good reliability and validity (Quittner & Espelage, 1999). However, the DPD method is still a form of self-report with other disadvantages including: the researcher having to be specially trained in its use; the time-consuming nature of collecting data; and the complex nature of the resulting information (Modi & Quittner, 2006).

Pharmacy databases record details of the type, quantity and dates of refills of medication dispensed and this information has also been utilised for the purposes of studying adherence in CF (Riekert, Mogayzel, Bilderback, Hale, & Boyle, 2007). In identifying those patients who refill and fail to refill their prescriptions, a global estimate of adherence and non-adherence can be calculated for research purposes. However, whilst this may provide accurate information for classifying those patients who fill in and collect their repeat prescription, pharmacy data only circuitously indicates whether medications were consumed or taken as directed (Myers & Midence, 1998; Briesacher, Andrade, Fouayz, & Chan, 2008). In addition, they may fail to capture those medications dispensed directly within hospital settings (Modi et al., 2006).

Electronic monitoring devices (EMDs) which utilise microchip technologies are considered a more objective and valid method of measuring adherence (Bender, Milgrom, & Apter, 2003). In studies of CF, several EMDs have been used to measure adherence including

Medication Event Monitoring System (MEMS) caps (i.e., a cap which records medication bottle openings) and inhalation devices (Latchford, Duff, Quinn, Conway, & Conner, 2009; McNamara, McCormack, McDonald, Heaf, & Southern, 2009; Dziuban, Saab-Abazeed, Chaudhry, Streetman, & Nasr, 2010). Although EMDs can provide an accurate and detailed measure of the date and time of treatment behaviour, there are certain difficulties associated with specific devices. MEMS caps, for example, measure the frequency of the medication bottle being opened but do not necessarily measure whether the medication has been taken or not (Myers & Midence, 1998). Although considered more valid, the results from studies utilising nebulisers may still be confounded due to inaccurate device technique and the phenomenon of “dumping” (i.e., administering and discarding excessive doses of medication) (Otsuki, Clerisme-Beaty, Riekert, & Rand, 2008).

The advent of Adaptive Aerosol Delivery™ (AAD) technology – which only electronically records nebuliser use on breath inhalation – provides a useful solution to “dumping” and progresses towards offering a “gold standard” measure of adherence (Duff & Latchford, 2010a). McNamara and colleagues (McNamara et al., 2009) utilised one such device (I-neb™, Respironics, Chichester, UK) in a recent study designed to determine adherence to aerosol therapy in a paediatric CF population. The I-neb™ device has a built-in Patient Logging System (PLS) capability which consists of a memory chip that can record up to 4000 treatments. PLS data from the I-neb™ was downloaded and used to calculate morning, evening and overall monthly adherence to aerosolised antibiotic therapy for over a year in a sample of children and adolescents infected with *Pseudomonas aeruginosa* (a naturally occurring bacteria, which if chronically established can lead to greater symptom severity and rapid decline in lung function for patients with CF) (Thomson & Harris, 2008). Although overall monthly adherence was maintained between 60% and 70% over the year, considerable variation both between and within participants was demonstrated and adherence was found to be better in the evening than it was in the morning. There are a number of potential explanations for these findings, but in the absence of detailed qualitative information exploring the experiences of adhering to aerosol therapy, further elucidation is not possible.

For patients with CF, aerosolised treatments are used almost universally. Newer AAD devices (such as the I-neb™) use electronic technology to adjust the timing of aerosol delivery to the patient’s individual breathing pattern. This helps to counteract some of the difficulties

associated with older, continuously operating nebuliser devices where it was common for over half of the drug to be wasted while the patient exhales (Kesser & Geller, 2009). Unlike the older, conventional nebulisers, the I-neb™ device is battery operated, silent, smaller and more portable. The liquid-crystal-display screen incorporated into the I-neb™ provides the user with continuous feedback on the device functions along with more tactile patient feedback (i.e., the use of a cascading vibration system) at the end of a completed treatment (Denyer & Dyche, 2010). Denyer and Nikander (2010) suggest that “the combination of a high lung deposition, minimal loss of aerosol during exhalation, and short nebulisation times makes the I-neb AAD System especially valuable to patients that are on multiple daily nebulisation treatments, are using drugs that should not be wasted into the room air, or would benefit from a more efficient delivery system” (pg.57). Indeed, studies investigating the acceptability of the device have found that the majority of patients rated the I-neb™ as either *easy* or *very easy* to use (Denyer, Black, Nikander, Dyche, & Prince, 2010; Denyer, Prince, Dixon, Agent, Pryor, & Hodson, 2010). The PLS capability provides additional feedback and can be actively utilised by clinicians to objectively monitor their patients and identify those who are not adhering to their aerosol therapy treatment (Denyer & Dyche, 2010). However, despite the technological advances in systems used to deliver aerosol therapy treatments and the greater patient acceptability of these devices, adherence rates remain relatively low (Latchford et al., 2009; McNamara et al., 2009). This is problematic given the prominence of aerosolised treatments in current CF drug-delivery regimens and the likely increase of their use in the future (Cystic Fibrosis Foundation, 2010; Duff & Latchford, 2010a).

1.4.1.2 Correlates of adherence to treatment in CF

Although studies have only recently begun to examine adherence difficulties to specific components of the treatment regimen in CF (e.g., Modi & Quittner, 2006), more general research on barriers to adherence has attempted to gain a greater understanding of what makes treatment problematic for patients and their families. Much of this research has focussed on the adult CF population (e.g., Abbot et al., 1994; Conway, Pond, Hamnett, & Watson, 1996; Abbot & Gee, 1998) and echoes the more general situation outlined earlier regarding chronic health condition studies investigating adult as opposed to child and adolescent adherence.

However, the last decade in particular has seen an increase in the number of research studies examining adherence within the child and adolescent CF population. It has been consistently demonstrated in CF for example, that age plays a significant role; adherence decreases as children transition to adolescence with the poorest rates occurring at around the age of 16 (Quittner, et al., 2000; Zindani et al., 2006; Rieckert, et al., 2007). Foster and colleagues (Foster et al., 2001) suggest that parental involvement in early childhood may explain higher levels of adherence as do Modi et al (Modi, Marciel, Slater, Drotar, & Quittner, 2008) whose research demonstrated better adherence for both preadolescents (ages 8 – 11) and adolescents (ages 11 – 16). These findings contrast with Drotar and Ievers (1994), however, who found that increased parental involvement was associated with lower levels of adherence. The role of the family does appear to be important, with one longitudinal study demonstrating how family dysfunction can discourage optimal treatment adherence and, in turn, negatively impact on CF health outcomes (Patterson, Budd, Goetz, & Warwick, 1993). In contrast, family cohesion and positive social support has been associated with better adherence to treatment both within chronic illness and within CF (Eddy, Carter, Kronenberger, Conradsen, Eid, Bourland, & Adams, 1998; Foster et al., 2001; DiMatteo, 2004). The finding that parents and the family may be an important factor associated with adherence to treatment in CF is perhaps unsurprising when considering Bronfenbrenner's (1977) Ecological Systems Theory. A core principle of this theory is the notion of *reciprocity*, both between individuals and their environment and between individuals within the system. The system clearly extends beyond the immediate family unit and so the finding that peer support for both CF patients and parents is thought to be an important factor associated with improved adherence (Barker, Cohen, Driscoll, Modi, Quittner, 2008) is again not surprising.

A lack of treatment specific knowledge and misunderstandings regarding treatment requirements have been identified as further contributors to poor adherence in CF (Koocher, McGrath, Gudas, 1990; Modi & Quittner, 2006). Ievers-Landis and colleagues (Ievers-Landis, Brown, Drotar, Caplan, Pishevar, & Lambert, 1999) reported these gaps and misunderstandings to be as high as 33% for mothers with school-aged children.

Perhaps unsurprisingly, the time consuming and complex nature of the treatment regimen in CF has also been shown to contribute significantly to poor adherence in children and adolescents (Modi & Quittner, 2006; Williams, Mukhopadhyay, Dowell, & Coyle, 2007; Bucks,

Hawkins, Skinner, Horn, Seddon, & Horne, 2009; Bregnballe, Schiøtz, Boisen, Pressler, & Thastum, 2011). Linked to the problem of time-management is the issue of forgetfulness (both intentional and unintentional) which has been found to act as another barrier to optimal adherence (Modi & Quittner, 2006; Dziuban et al., 2010; George, Rand-Giovannetti, Eakin, Borrelli, Zettler, & Rieker, 2010; Bregnballe et al., 2011).

In examining adherence in children and adults with CF, Arias Llorente and colleagues (Arias Llorente et al., 2008) reported that treatment adherence decreases as the disease becomes more severe. These results are in contrast to other studies however, which demonstrate an increase in treatment adherence with severity of CF (Michaud, Frappier, & Pless, 1991; Zindani et al., 2006). In attempting to explore the relationships between illness perceptions, emotional representations, treatment beliefs and reported adherence in CF, Bucks and colleagues (Bucks et al., 2009) found that adolescents reported doubts about the necessity of chest physiotherapy and antibiotics. Along with unrealistic beliefs about the chronicity of CF, these treatment beliefs were significant predictors of poor adherence. The authors argue that their findings provide preliminary support for components of Leventhal's Self-Regulatory Model (Leventhal, 1993; Leventhal et al., 1984) in explaining adherence in adolescents with CF. They do express caution in interpreting the results of their study however, not least because of the problems highlighted above in relying on self-report to measure adherence outcomes.

1.4.1.3 Interventions to improve adherence to treatment in CF

A better understanding of the factors affecting adherence has enabled a limited number of interventions – designed to increase adherence to treatment in the CF population – to be developed and studied. Stark et al (Stark, Quittner, Powers, Opiari-Arrigan, Bean, Duggan, & Stallings, 2009) for example, found that a combined group behavioural and nutrition education intervention, that involved both parents and children successfully increased adherence to dietary intake in children with CF. In their systematic review, Bernard & Cohen (2004) found that behavioural techniques (e.g., behavioural modification and token economies) can increase adherence to diet and chest physiotherapy in CF. However, the authors make a number of recommendations for future research and in particular suggest that CF research should diversify away from an emphasis on studies examining Caucasian females in the preadolescent age group. At present, there are no published studies that have examined the effectiveness of interventions

specifically designed to improve adherence to aerosol therapy (Duff & Latchford, 2010b). Again, as highlighted earlier, this is somewhat concerning when taking into account the importance of this type of therapy in the CF treatment regimen.

1.5 Rationale for the current study

Notwithstanding the difficulties associated with measurement, it is clear from examining the literature that adherence to treatment in CF is generally poor, and subject to the influence of a number of interacting factors, particularly in children and adolescents where the context of the family adds a further layer of complexity. Barker & Quittner (2010) have attempted to discuss these complex interactions by proposing a developmental, biopsychosocial model of the disease that “highlights the bidirectional influences of age-related development on disease management and progression” (pg. 468). Psychological and social factors are clearly important with Dziuban et al (2010) suggesting that in addition to the findings described above, other obstacles to treatment adherence “may lie deeper in the psychosocial framework” (pg. 456). This may well be true, with one study finding that parents were actually unable to identify what was hindering their child’s adherence to aerosolised therapies perhaps suggesting the possible influence of deeper, less conscious processes (Modi & Quittner, 2006).

Technological advances in systems and devices designed to administer aerosol therapy have resulted in treatments becoming quicker and more effective. However, studies utilising intelligent data downloaded from these devices indicate that adherence rates to aerosol therapy remain low. The absence of detailed qualitative information exploring the experiences of adhering to aerosol therapy has resulted in potential explanations for these findings being difficult to ascertain. In addition, while much has been achieved in recent years in terms of interventions designed to treat adherence problems and thus maximise treatment potential (Bernard & Cohen, 2004; Kahana, et al., 2008); gaining a more detailed understanding of the factors influencing adherence to specific components of the CF treatment regimen may be essential if successful interventions are to continue being informed and further developed. This is true for aerosol therapy, particularly within the adolescent CF population where adherence is low but fewer interventions have been specifically targeted (Duff & Oxley, 2007; McNamara et al., 2009; Duff & Latchford, 2010b).

The above discussion highlights the need and value of utilising qualitative methodologies in attempting to uncover the likely, deep-rooted, multi-factorial influences on adherence to specific components of the treatment regimen within the adolescent CF population.

1.5.1 Introducing the current study

With the influence of parents and the family clearly being an important factor associated with adherence in adolescents, a need exists for a richer and more detailed understanding from both a patient and a parent perspective as to the process of adherence to treatment in CF. The paucity of research examining adherence to specific components of the treatment regimen, particularly within the adolescent CF population, makes it especially important to gain this more focussed understanding. With the prominence of aerosol therapy in current CF drug-delivery treatment regimens and the likely increase of their use in the future, it seems prudent to prioritise this as an area of enquiry. Further benefits lie in the ability of AAD devices to provide an objective and accurate measure of adherence to aerosolised therapy treatments thus enabling accounts of adherence to be anchored in everyday life and understood in the context of day-to-day behaviour.

The current study, therefore, was designed to elicit the perspectives of adolescent patients and their parent on adhering to aerosol therapy. In order to reduce potential bias and maximise the potential for obtaining rich and detailed accounts, it was decided to use qualitative methods and to interview patient and parent about their experiences of adherence over recent weeks. To provide a cue to participants about the actual rates of adherence, and to prompt discussion of particular events, the ability of the I-neb™ device to produce data on its use was utilised to provide a diary for each participant showing actual rates of adherence day-by-day for the previous week. It is hoped that the findings of the study will complement and extend the previous research on adherence to treatment in CF and help to inform clinical and research practice.

1.5.2 Research questions

- How is the process of adhering to aerosol therapy experienced by both an adolescent patient and a parent?
- What are the differences and commonalities in the experience of aerosol therapy compared to the other therapies used in CF?

- Does the use of PLS data downloaded from the I-neb™ device help to qualitatively explore the process of adhering to aerosol therapy?

1.5.3 Research aim

To qualitatively explore the process of adhering to aerosol therapy from both an adolescent patient and a parent perspective and to utilise a seven-day retrospective adherence data download taken from the I-neb™ device in order to further elucidate this process.

2.0 METHOD

2.1 Design

A qualitative research design employing semi-structured, face-to-face interviews was used for the purpose of this study. The semi-structured interviews were conducted with six adolescent CF patients and their parent (n=12) on an individual, separate basis in an attempt to illicit participants own experiences, understandings and perspectives of the process of adhering to aerosol therapy treatment. In order to anchor experiences in real life examples, part of the interview was guided by a seven-day retrospective printout of the Patient Logging System (PLS) data downloaded from the I-neb™ device. The data generated from the interviews were transcribed and then analysed using the Grounded Theory Method (GTM). The systematic coding of the data enabled the emergence of core categories which were then structured and presented as a separate theoretical formulation which represented both the parent and adolescent experiences of the process of adhering to aerosol therapy treatment.

2.2 Methodological considerations

This study used the GTM as the overarching approach to data collection and analysis. The following section will briefly outline the purpose of qualitative research, describe the GTM method and provide a rationale for its use.

2.2.1 Qualitative research

Qualitative research seeks to provide rich and detailed descriptive accounts of the phenomenon under investigation (Geertz, 1973; Ritchie & Lewis, 2003; Smith, 2008). In psychology, qualitative research is primarily concerned with accessing and exploring participants' own unique meanings attached to their behaviour, thoughts and feelings (Willig, 2008). The ability of a qualitative approach in “exploring, describing and interpreting the personal and social experiences of participants” (Smith, 2008, pg. 2) positions it as a particularly useful methodology in attempting to address the main aim of this study.

Qualitative research however, is not a homogenous speciality. There are a variety of qualitative approaches that gather under the umbrella term of “qualitative methodology”. Methods such as grounded theory (Glaser & Strauss, 1967), interpretative phenomenological

analysis (IPA: Smith, 1996) thematic analysis (Braun & Clark, 2006) and discourse analysis (Potter & Wetherell, 1987) each have their own theoretical and methodological commitments. Although diverse in their application, there are some important recurrent themes within all qualitative research methodologies which suggest that significant similarities exist. Miles and Huberman (1994) summarise these recurrent features which include: the commitment of qualitative research to study phenomenon in naturalistic settings; the fact that the method is inductive, with the participant often leading the process of data generation; the fact that most analyses are conducted on words (e.g., written accounts or interview transcripts); and the fact that the researcher is viewed as an “instrument” within the study who plays a central role in making sense of and interpreting the phenomenon under investigation.

Whilst a number of qualitative approaches are available, it will be argued that the GTM is the most suitable approach for the purpose of this research.

2.2.2 Description of the Grounded Theory Method

The term “Grounded Theory” relates both to the result of an overall methodological approach to research (i.e., a way of developing a theory that has been generated and is therefore “grounded” in data) and a specific set of methods, principles and procedures used for data analysis that was first advocated by sociologists Barney G. Glaser and Anselm L. Strauss in the 1960’s (Henwood & Pidgeon, 2006). Although the above distinction may appear relatively straightforward, this explicit difference between “methodology” and a “set of methods” is often absent within the literature and can lead to confusion, particularly for those relatively new to the subject (Charmaz, 2008).

Qualitative methods were viewed with suspicion in the 1960’s and criticised for being unsystematic, anecdotal and biased (Charmaz, 2006). Glaser and Strauss’s seminal text – *The Discovery of Grounded Theory* (1967) – is often described as “cutting-edge” because it challenged that view and the dominance of sophisticated quantitative methodologies that were rooted in positivism (i.e., assuming a belief in scientific objectivity, logic and truth) and very much favoured by the scientific community at that time (Charmaz, 2008). The book aimed to legitimise rigorous qualitative research and support its own intrinsic value and integrity that was distinct from quantitative research (Bigus, Hadden, & Glaser 1982). Denzin & Lincoln (1994)

argue that their book was a key force that helped to spearhead the “qualitative revolution” that in the latter part of the twentieth century was gaining momentum.

Since the publication of *The Discovery of Grounded Theory* (1967) the authors have seemingly evolved and developed the GTM in divergent directions; Glaser (1978) arguing for a strict positivist approach to data with the researcher remaining entirely objective compared to Straus who, along with later collaborator Juliet Corbin (Straus & Corbin, 1990), suggested that data needed to be reduced into more manageable forms with some level of researcher interpretation a necessity. Further diversification occurred with the development of Charmaz’s (1990; 2000) Constructivist GTM. Her version emphasises how “data, analysis, and methodological strategies become constructed, and takes into account the research contexts and researchers’ positions, perspectives, priorities, and interactions” (Bryant & Charmaz, 2007, pg. 10). Although debates as to the relative merits of a Glaserian, Straussian or Constructivist approach continue (e.g., Boychuk-Duchscher & Morgan, 2004), some argue that dividing the GTM into these rigid categories is not particularly helpful (Annells, 1996; Birks & Mills, 2011).

Heath and Cowley (2004) agree and suggest that researchers – particularly those new to the GTM – should select the approach that best suits their style and enables them to achieve an appropriate and useful grounded theory. Birks & Mills (2011) concur with this position and argue that methodologically “there are no right or wrong approaches to using grounded theory methods” (pg. 8). Bryant and Charmaz (2007) have suggested that instead of viewing grounded theory methods as detailed, rigid prescriptions for research practices and procedures, researchers should treat them as heuristics and guidelines which provide *tools to use* as opposed to *recipes to follow*.

2.2.3 Rationale for using the Grounded Theory Method

In considering some of the background, debates and processes associated with the GTM, one can appreciate its position as a popular and useful qualitative approach to research. Birks & Mills (2011) argue that an integrated and comprehensive grounded theory that explains a process associated with a given phenomenon is the final product of a grounded theory study. They suggest that it is suitable to use when little is known about the area of study and when the desired outcome is the generation of a theory with explanatory power. It is for these main reasons that the GTM has been selected as the most appropriate to utilise for the purpose of this study.

Although other qualitative methodologies were considered – in particularly IPA (Smith, 1996) and thematic analysis (Braun & Clark, 2006) – they did not enable the development of a theoretical formulation which assisted in enhancing the understanding of the process of adhering to aerosol therapy from a patient and parent perspective. Furthermore, the chief investigator has a preference for the GTMs emphasis on taking a systematic, step-by-step approach to analysing data. As the chief investigator had prior knowledge of the adherence literature, this systematic approach was particularly helpful in reducing potential bias.

2.3 Ethical Considerations

2.3.1 Ethical Approval

The study was originally reviewed by an independent academic panel at The University of Leeds in November 2010. A full ethics application was then submitted to the NHS National Research Ethics Service (NRES). This was reviewed by the NRES Yorkshire and the Humber committee in July 2011. Following the satisfactory completion of some minor amendments, the study was granted ethical approval on 12th September 2011 (see Appendix 1). The amendments included some minor details being added to the participant information sheets, the inclusion of a standardised letter to be sent to the participants' general practitioner and for boxes that enabled the participants to initial each question answered to be added to the consent form. NHS Research and Development approval was obtained from the Leeds Teaching Hospitals NHS Trust on 10th October 2011 and from Alder Hey Children's NHS Foundation Trust on 17th September 2011.

2.3.2 Informed Consent

It was important that the participants were able to give informed consent to take part in the study. Participants were approached by the CF specialist physiotherapist during routine outpatient appointments, not the chief investigator. Interested participants were provided with information sheets appropriate to their age. One was for those participants aged 11 to 13 (see Appendix 2), one was for those aged 14 to 16 (similar to Appendix 2), and one was for the parent (see Appendix 3). Participants had the opportunity to ask questions about the study at this time and again when the chief investigator contacted them. If they agreed to take part, then the written consent form (see Appendix 4) was completed at their follow-up outpatient appointment.

2.3.3 Anonymity

The dignity, rights, safety and well-being of participants was an absolute priority throughout the process of conducting the research. This included the assurance of anonymity. Participants were informed that the seven-day retrospective PLS data downloaded from the I-neb™ device would be fully anonymised and sent via secure e-mail correspondence by the specialist CF physiotherapist to the chief investigators NHS Net mail account ahead of the semi-structured interview. In addition, an I.D. number was given for each participant and used on all interview recordings and transcripts. Any identifiable person or place name was removed from the transcript. During the process of analysis, pseudonyms were used to ensure that any quotes selected to feature as part of the results would not easily identify the participants.

2.4 Sampling

The study used aspects of “purposeful sampling” (i.e., whereby the researcher actively seeks participants who offer a rich source of information that will likely help to meet the overall research aim) in that only those who had experience of using the I-neb™ device for at least the last 12 months as part of their treatment regimen were invited to take part. However, the CF physiotherapist approached these potential participants in order of their attendance at routine appointments to reduce bias. In this way, a selection of patients typically encountered within routine clinical practice (and their parents) was selected to take part in the study.

2.4.1 Participants

Participants were adolescents and parents recruited from two regional paediatric CF centres: Leeds General Infirmary (LGI) and Alder Hey Children’s Hospital (AHCH), Liverpool. A total of six adolescent participants aged 11 – 16 were recruited (three from each centre). Parents of the participants were also recruited, resulting in a total sample of N=12. Those identified as the primary caregiver, most commonly mothers (Timko, Stovel & Moos, 1992) were invited to take part. Adolescents were eligible for participation if they were 11-16 years of age, had a confirmed diagnosis of CF via sweat testing and had been using the I-neb™ device for at least the last 12 months as part of their treatment regimen. Participants were excluded from the study if they were under 11 years or over 16 years of age. They were also excluded from the study if they were not deemed clinically stable (i.e., if they had had an FEV1 reading <50% over the last 12 months).

2.5 Recruitment of participants

After ethical approval was granted, a list of patient names who met the inclusion criteria was compiled by the specialist CF Physiotherapists at LGI and AHCH. Potential participants (adolescent patient and their parent) who met the inclusion criteria were approached about the study by the CF physiotherapists during their routine outpatient appointment. This was done on a *first-come, first-served basis* (i.e., the patients who had their outpatient clinics scheduled at the earliest date were the first to be approached). Interested patients and their parent received an information sheet (one for the patient using age appropriate language and one for the parent) regarding the study and were encouraged to ask questions. At this point, the next outpatient appointment was arranged for approximately 6 weeks time by the CF Physiotherapists as standard. Interested participants were informed that the chief investigator would telephone potential participants to answer any further questions they may have about taking part in the study and to arrange a convenient time for the potential interview to take place (i.e., within 7 days after the next outpatient appointment). The chief investigator also telephoned the participants the day before their next scheduled outpatient appointment to remind them both of the importance of bringing along the I-neb™ to their appointment and of the date and time of the pre-arranged semi-structured interview.

Participants attended their next scheduled outpatient clinic appointment as planned and were again provided with the original participant information sheet in order to reiterate that their PLS data routinely downloaded within the outpatient clinic appointment would be sent via secure e-mail correspondence by the specialist CF physiotherapist to the chief investigators NHS net e-mail account ahead of the semi-structured interview. Participants were then asked to sign a form which consented to their anonymised adherence data being sent to the chief investigator and which consented to them taking part in the study. The anonymised PLS data downloaded from the I-neb™ at the outpatient clinic appointment was then sent via secure e-mail correspondence. The chief investigator met the participants at the scheduled time in order to conduct the semi-structured interview. Participants predominantly requested that home interviews were conducted as these were the most convenient.

2.6 Data collection

2.6.1 Interview schedule

Two interview schedules were used; one for the adolescent patient (see Appendix 5) and one for their parent (see Appendix 6). Although both were similar, the interview schedule that was devised for the adolescent patient used language that was slightly easier and more appropriate for the participants' level of development. Each interview schedule was based upon the recommendations made by Charmaz (2006) and consistent of 11 open-ended questions that were simple, clear, brief, forthright and jargon-free (see Appendix 5 and 6). Each of the 11 questions had associated prompts that were designed to elicit full and rich accounts from the participants. The interview schedule for the adolescents was designed to elicit a description of the experience of adhering to aerosol therapy. For the parents, the schedule was designed to elicit a description of their experiences of their child adhering to aerosol therapy.

The development of the interview schedule took place over a number of weeks. Charmaz (2006 pgs. 30-31) provides a series of sample grounded theory interview questions and many of these were adapted and used to develop the initial interview schedule for the current study. The draft questions were discussed and revised during a pre-arranged supervision session. The resulting draft interview schedule was piloted on a trainee clinical psychology colleague and an adolescent family member, with the resulting suggestions for improvements in the phrasing of the questions being made. Further information about how the interview schedule was used alongside the Patient Logging System data is provided in section 2.6.3.

2.6.2 Patient Logging System data

The third generation AAD system nebulizer – the I-neb™ (see Figure 4) has a built-in Patient Logging System (PLS) capability which consists of a memory chip that can record up to 4000 treatments. Every nebulised dose of medication is recorded as an individual line of data and includes the time and date, drug code, dose delivered (either Full, <12.5%, or between >12.5% and <100%), breathing mode (Tidal Breathing Mode [TBM] or Target Inhalation Mode [TIM]) and duration of treatment. The I-neb™ device is able to transmit this recorded PLS data to a PC via an infrared interface.

It was the most recent seven days worth of this PLS data (downloaded as standard within the outpatient appointment) that was anonymised and sent via secure e-mail to the chief

investigator ahead of the pre-arranged semi-structured research interview. This information was then transferred onto a more easy-to-read sheet that resembled a seven-day diary (see Appendix 7). This information was presented to the parents and adolescents to assist them in reflecting on their recent experiences of adherence to aerosol therapy and to guide some of the questions on the interview schedule.



Figure 4: The I-neb Adaptive Aerosol Delivery (AAD) System

2.6.3 Semi-structured interviews

For their convenience, the participants decided who was to be interviewed first. Based on the guidelines suggested by Fylan (2005, pp. 69-74), the chief investigator attempted to establish and maintain rapport with participants throughout the interview process. The use of the interview schedules allowed sufficient structure to focus the interview although the use of “topic prompts” allowed for flexibility and ensured that an over-reliance on fixed questions was avoided. The order of questions was changed on an ad hoc basis depending on the specific idiosyncrasies of each interview. The PLS data was used to guide some of the open-ended interview questions which allowed participants to reflect on their recent experiences of adhering to aerosol therapy over the previous week. As the interviews progressed, it was noticeable that the chief investigator attempted to ensure that the PLS data was introduced earlier on in the

interview process as this was proving to offer valuable insight into the participants lived experiences and encouraged interesting reflections on specific adherence situations. The majority of interviews lasted approximately one hour each, and were all conducted in private.

2.7 Data analysis

The data analysis process was guided in particular by the models used by Straus and Corbin (1990) and Charmaz (2006). The coding process began with transcription of all the interviews which were then subjected to the structured process of open, axial and theoretical coding. These coding methods will be outlined in the following section along with a worked example to illustrate the process more fully. Data analysis was supported by a number of other techniques commonly used within the GTM including *constant comparison*, *memo writing* and *diagramming*. These techniques will also be outlined before moving on to describe the tools used to ensure that the overall quality of the data analysis process was maintained.

2.7.1 Transcribing

All of the digitally recorded interviews were transcribed by a trained third party. All recorded interviews and subsequent interview transcripts were password protected. The chief investigator listened back to all of the recorded interviews alongside the transcript to ensure accuracy and to enable familiarity with the participants' narrative accounts (Henwood & Pidgeon, 2006).

2.7.2 Coding

Bryant and Charmaz (2007) offer a very straightforward definition of coding in the GTM: "Coding is the process of defining what the data is about" (pg.605). In the GTM, this *defining of data* is done in a series of stages. The open coding process begins by analysing the transcripts line-by-line and assigning a code to each. Axial coding is a method of grouping these open codes into a series of emerging categories. Finally, theoretical coding attempts to identify the possible relationships between these categories which are then used as hypotheses to be integrated into a theoretical formulation of the overall process. The constant comparison of incidents within the data, incidents with categories, categories with other categories and categories with emerging theoretical propositions is a method used throughout the lifetime of the project. Memos of the researcher's thoughts (from initial and basic to more advanced and

theoretical) are used alongside coding and constant comparison with the explicit aim of taking conceptual development forward. Closely related is the use of diagrams which create a visual display that helps to conceptually map analysis throughout the entire process.

Before moving on to describe each of these methods in more detail, a diagram of the overall process outlined above is shown for the current study in Figure 5.

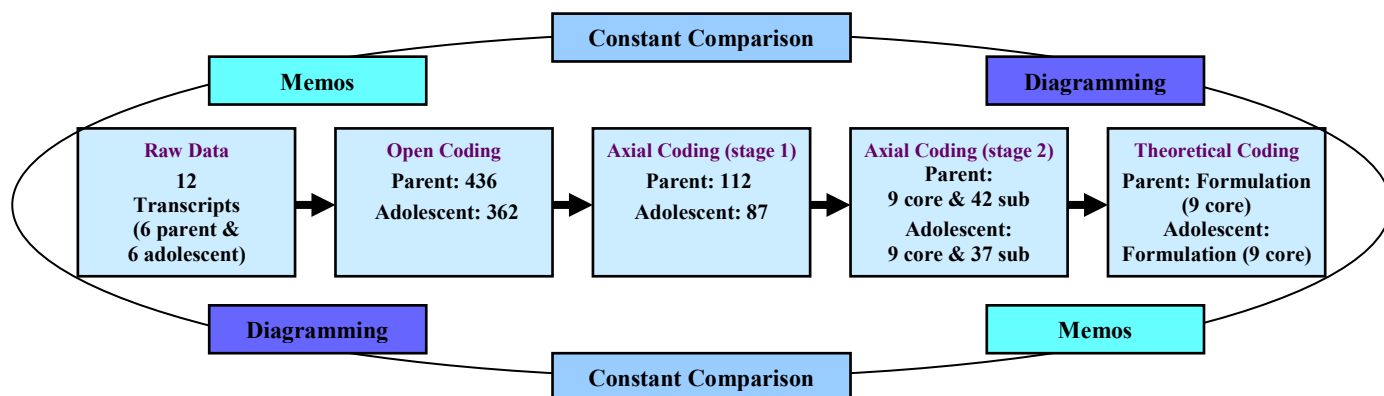


Figure 5: The GTM data analysis process

2.7.2.1 Open coding

The initial process of open coding is used to “fracture the data” (Glaser & Strauss, 1967) so as to compare incident with incident, name emerging patterns or apparent phenomena and instigate the practice of comparison between the codes applied. When coding, Charmaz (2006) argues for the use of gerunds (i.e., verbs used as nouns that always finish with ‘ing’) as a way of discovering process in the data in addition to focussing on the participant’s experiences as a source of conceptual analysis. In this way, codes are prevented from being used simply as a descriptive summary of an event; enabling the first level of interpretation to begin by assigning a *label* to a unit of meaning (Strauss & Corbin, 1990).

The chief investigator began the open coding process by working through a transcript, and assigning a code to each line of text. Glaser (1978) poses three questions to be asked of the data, with Charmaz (2006) adding a fourth: what is this data a study of?; what category does this incident indicate?; what is actually happening in the data?; and from whose point of view? The chief investigator utilised these questions throughout which helped to preserve the data analysis process and allowed different theoretical possibilities to remain open (Charmaz, 2006). Although parent and adolescent transcripts were analysed separately, they were also analysed concurrently (i.e., a parent transcript was analysed and then an adolescent transcript was

analysed and vice versa). This enabled the process of constant comparison both within and between transcripts and helped to prevent the forcing of data into a preconceived framework (Glaser, 1992). The line-by-line open codes for each transcript were written on post-it notes (with the associated page number) and transferred to the back pages (see Appendix 8). The chief investigator regularly returned to the open codes, using the above questions to ensure that the meaning captured was accurate. Those codes that did not were re-categorised or removed. The initial stage of open coding produced 436 codes for the parent analysis and 362 codes for the adolescent analysis. An example of the open coding process is shown in Table 1.

Quote from participant	Example of open coding
<i>Parent 3: "I like to know she's done it [aerosol therapy] because if she hasn't done it I worry and again it's, she'll say I'm a control freak and I probably am a bit but having had all the years where I had complete control over her treatments to almost having no control at all now, erm yeah I like to remind her, just check in my own mind that she's done it.</i>	Monitoring adherence Feeling worried Child's frustration Behaviour has an impact Having control over treatments Feeling out of control Supervising strategy Reminding reduces parental anxiety

Table 1: Example of the open coding process

2.7.2.2 Axial coding

Data analysis continues with axial coding, which was described by Strauss and Corbin (1990, pg.96) as "a set of procedures whereby data are put back together in new ways after open coding, by making connections between and within categories". Stage one of axial coding is where open codes are compared and reviewed both within and between transcripts in order to find similarities in meaning. In the current study, all of the generated open codes were written onto the back of each transcript (see Appendix 8) to enable this comparison to take place.

Strauss & Corbin (1998) suggest that similar codes are then grouped together into emerging subcategories that begin to form the initial analytic framework. These axial code subcategories (and associated open codes) in the current study, were written onto the back of each transcript (see Appendix 9). At this stage, 436 open codes for the parent data were reduced

to 112 axial code subcategories. For the adolescent data, 362 open codes were reduced to 87 axial codes.

The second stage of axial coding aims to link the subcategories to the emerging core categories and asks how they are related (Charmaz, 2006). In the current study, this was achieved by transferring each axial code subcategory onto a post-it note and arranging them diagrammatically on large pieces of card (see section 2.8.3). Memo writing was used alongside this process to capture analytical thoughts and to help drive the development of theoretical ideas (see section 2.8.2). Strauss and Corbin (1990) advocate the use of diagrams such as these from the commencement of a study in tandem with the writing of memos. In this second stage, 112 axial code subcategories for the parents were reduced and organised hierarchically underneath 42 final subcategories and 9 core categories. For the adolescents, 87 axial code subcategories were reduced and organised into 37 final subcategories which sat under 9 core categories.

The process of regular supervision at this stage helped to provide a more objective view of the emerging conceptual organisation of subcategories. To provide an illustrative example of the progression from open to axial coding, the open codes from Table 1 have been used to show how the coding process developed (see Table 2).

Open code	Axial code subcategory
Monitoring adherence	Reminding
Feeling worried	Anxiety and worry
Child's frustration	Child feeling emotional
Behaviour has an impact	Responsibility, independence and the developing child
Having control over treatments	Feelings of control
Feeling out of control	Feelings of control
Supervising strategy	Reminding
Reminding reduces parental anxiety	Reminding / Anxiety and worry

Table 2: Example of the axial coding process

2.7.2.3 Theoretical coding

Although the core categories and subcategories had been developed, they lacked an overall theoretical structure. Glaser (2005) suggests that this is not uncommon and argues that

theoretical coding, although difficult, helps to form a theory which explains how categories relate to each other. He goes on to say that theoretical codes can be drawn from existing theories to assist in theoretical integration. In using this method, Birks and Mills (2011) suggest that “explanatory power is added to the final product of a grounded theory study by situating it in relation to a theoretical body of knowledge.”

In the current study, the post-it note diagrams were used alongside theoretical memos to arrange the core categories into a coherent story which best reflected the experiences of the participants. This enabled the development of a theoretical formulation which described the process of adhering to aerosol therapy from both a parent and adolescent patient perspective. Table 3 provides an illustrative example of how the open and axial code subcategories relate to the core categories and processes described in the theoretical formulation for the parent data.

To reduce subjectivity, research supervision was used to check that the theoretical formulation best represented the participants’ narrative accounts. This enabled the formulation to be openly discussed and refined.

2.8 Other procedures in the methodology

2.8.1 Constant Comparison

As previously highlighted, constant comparison is an important feature of the GTM and was used throughout the process of analysis in the current study. Bryant and Charmaz (2007) define constant comparison as a “method of analysis that generates successively more abstract concepts and theories through inductive processes of comparing data with data, data with category, category with category, and category to concepts” (pg.607).

At the level of open coding, parent and adolescent data were compared both within and between transcripts and coded repeatedly, enabling any new codes to be checked against existing ones. At the level of axial coding, constantly comparing potential subcategory codes enabled them to be checked to see if they were describing the same phenomena. At the level of theoretical coding, comparing the emerging theoretical formulation with the participant accounts within the raw data helped to ensure that the resulting grounded theory best represented the experiences of the parents and adolescents.

Table 3: Example of the overall data analysis coding process

Open code	Axial code subcategory	Core Category	Process described
Monitoring adherence	Reminding	Strategies and behaviours	Cognitive, affective and behavioural
Feeling worried	Anxiety and worry	Emotional drivers	Cognitive, affective and behavioural
Child's frustration	Child feeling emotional	Parent's experience of their child's adherence behaviour	Parent's interaction with their child
Behaviour has an impact	Responsibility, independence and the developing child	Parent and adolescent relationship	Parent's interaction with their child
Having control over treatments	Feelings of control	Emotional drivers	Cognitive, affective and behavioural
Feeling out of control	Feelings of control	Emotional drivers	Cognitive, affective and behavioural
Supervising strategy	Reminding	Strategies and behaviours	Cognitive, affective and behavioural
Reminding reduces worry	Reminding / Anxiety and worry	Strategies and behaviours and Emotional drivers	Cognitive, affective and behavioural

2.8.2 Memo writing

Memos have been described by Clarke (2005) as “intellectual capital in the bank.” Memos are a written record of a researchers thinking throughout the process of undertaking a GTM study. The chief investigator found that numerous memos were stimulated during the different stages of coding. Henwood & Pidgeon (2006) suggest that the content of memos should not be constrained in any way and can include hunches and insights, comments about specific codes and categories, deliberations about refinements and explanations of modifications to categories. Memos were kept on post-it notes and used to record exactly the types of content recommended above. There were used as both a helpful memory aid during the analytic process and as an essential tool in the development of the overall theoretical formulation. An example of a memo is shown in Figure 6. In addition to using memos, the researcher kept a reflective research journal which was used primarily to capture thoughts immediately after conducting the semi-structured interviews. These journal entries were a valuable source of data when compiling the participant pen portraits (see section 3.2).

Memo date: 13.04.12

Parent 3: “I like to know she’s done it because if she hasn’t done it I worry and again it’s, she’ll say I’m a control freak and I probably am a bit but having had all the years where I had complete control over her treatments to almost having no control at all now, erm yeah I like to remind her, just check in my own mind that she’s done it” (Page 21: 907-913)

Anxiety and worry is present and appears to be driving reminding behaviour? Her daughters account suggest that reminders can help but are frustrating (pg. 13 356-562)

Figure 6: Example of a memo

2.8.3 Diagramming

Strauss and Corbin (1990) advocate the use of diagrams from the beginning of a study in tandem with the writing of memos. Birks and Mills (2011) suggest that diagramming whilst concurrently analysing data enables a greater sense of organisation of ideas. They go on to say that although initial coding may result in messy and intricate diagrams, more neat and simple diagrams will evolve as the researcher moves through axial and theoretical coding

stages. Diagramming was used in the current study throughout the three main stages of analysis. The fact that axial codes were kept on small post-it notes enabled large A1 size card diagrams to constantly evolve throughout the coding process. The process of diagramming was particularly helpful during axial and theoretical coding as a way of visually representing how the categories related to each other and how the processes described by the participants interacted with one another. Appendix 10 shows a picture of a diagram used during the axial coding stage of the parent data analysis.

2.9 Quality control

2.9.1 Supervision

Regular research supervision was arranged throughout the entire study. Supervision was particularly helpful during the process of data analysis. The systematic and visual approach adopted during data analysis enabled supervision to be used in a variety of ways to ensure adequate quality controls were in place. This included, for example, the checking of line-by-line initial codes, making comments and observations about axial codes and ensuring that the theoretical formulation could be further developed and refined. Minutes of the research supervision sessions were kept so as to provide a record of detailed discussions. This approach enabled the maintenance of an “audit trail” which Burns (1989) argues is a major factor in ensuring methodological and procedural rigour. Furthermore, being able to bring examples of the A1 sized card diagrams and copy’s of memos into supervision helped the research supervisor to make comments and suggestions that were based upon a visual representation of the chief investigators thought processes throughout the various stages of data analysis.

The chief investigator also attended a peer supervision group facilitated by an academic supervisor with decades of experience in qualitative methodologies. The peer supervision group provided additional support and helped to ensure that the data analysis process could be openly discussed and commented upon.

Although the use of supervision helped to reduce subjectivity, it is recognised that some form of researcher interpretation of the data is inevitable. This is discussed in more detail in section 2.9.3.

2.9.2 Consideration of other methods of quality control

There were other methods of quality control that were considered for use within the current study including *inter-rater reliability* and *respondent validation*. Although some

argue that estimates of inter-rater reliability (i.e., whereby different observers attempt to replicate the process of coding and calculate a percentage of agreement) should be included in qualitative studies (e.g., Lincoln & Guba, 1985) others suggest that the method is inappropriate for qualitative inquiry because the concept is based on positivist assumptions (Bogdan & Biklen, 1992; Marshall & Rossman, 1995). Goodwin and Goodwin (1992) further argue that although reliability and validity are relevant to qualitative research, “empirical estimation is difficult to impossible” (pg.416) and make concepts which rely on these techniques (such as inter-rater reliability) meaningless, particularly when one considers the ontological and epistemological foundations of qualitative methodology. Although estimates of inter-rater reliability were considered as a method of quality control in the current study, it was decided that efforts would be directed at ensuring adequate methodological and procedural rigour was maintained as opposed to utilising a technique that may not prove to be helpful or appropriate.

Respondent validation (i.e., whereby participants are asked to comment upon and critically analyse the coding process and/or the overall study findings) has received support as a helpful method of ensuring the overall validity of qualitative research (Smith, 2008). However, time constraints and the disparate geographical locations of the chief investigator and the participants did not make this technique a viable proposition.

2.9.3 Potential researcher bias

Numerous commentators have highlighted the need for researchers to be aware of their own potential biases and the way in which these can influence the overall research process (Bryant & Charmaz, 2007; Smith, 2008). Birks and Mills (2011) argue that the use of memos, reflective writing and the overall maintenance of an audit trail promote reflexivity and help the researcher to remain alert to the influence of potential biases. These techniques were used throughout the study and have been described in detail above. In addition, the following section will provide a brief description of the chief investigator and potential sources of bias. These biases are further considered in section 4.8.1.

2.9.3.1 Introducing the chief investigator

The chief investigator is a 35 year old, white, male psychologist in clinical training from Northern Lincolnshire. Prior to entering clinical psychology training, he worked within a large public health department where he became interested in health behaviour research and the wider determinants of health. These interests led to further academic study and he

completed an MSc in Health Psychology in 2009. Although he had prior knowledge of the adherence literature, this understanding was not in-depth and he was keen to delay a thorough review of the existing literature (so as to prevent existing theories and knowledge impacting on the study processes and outcomes) until after the data analysis was complete. While this was not entirely possible, due to the submission of an earlier researcher proposal, he returned to the literature to review this more thoroughly following the completion of the results chapter.

The chief investigator's recent clinical work has focussed on providing psychological support for adult patients with chronic physical health conditions such as heart disease, cancer and diabetes. He takes a person-centred approach to his clinical work and is influenced (and utilises) the ideas in cognitive behavioural, psychodynamic and systemic approaches. Prior to beginning the research study, he had never met anyone diagnosed with CF. He reflected both in his research journal and within supervision about his feelings towards the parent and adolescent participants. These primarily focussed on the admiration felt towards the participants for coping with the very real demands of living with CF. He also felt very privileged to hear the participants' accounts and was humbled by their experiences. This led to a passion for the research and a hope that the study findings would add to the existing research on adherence in CF.

The chief investigator has conducted a number of previous qualitative studies but had no prior experience of using the GTM.

3.0 RESULTS

3.1 Sample

12 participants (six child-parent dyads) took part in this study. The adolescent participants were all between the ages of 11 and 16 years. The parents taking part were predominantly mothers, with only one of the participants being a father. As explained in the methodology section, it was anticipated that more mothers than fathers would respond to the invitation to take part. Table 4 provides basic demographic details of the child-parent dyad participants along with information relating to the child's aerosol therapy regimen. Pseudonyms have been given to each participant in order to maintain confidentiality.

Table 4: Participant demographic information (N=12)

Participant*	Gender	Age at interview	Number of aerosolised treatments prescribed	Average 7-day adherence rate (%)	Average 7-day doses fully administered (%)	Mode of administration**	Average treatment administration time (minutes)
Karen	Female	16	1	28.57	14.29	TBM	10
Mary	Female	43	-	-	-	-	-
Tom	Male	11	1	100	100	TIM	1
Pippa	Female	39	-	-	-	-	-
Amanda	Female	14	1	71.43	42.86	TIM	12.2
Carla	Female	45	-	-	-	-	-
Elizabeth	Female	13	2	100	92.86	TIM	2.64
Greg	Male	46	-	-	-	-	-
David	Male	12	2	85.71	85.71	TIM	1
Lindsey	Female	37	-	-	-	-	-
Simon	Male	13	2	92.86	92.86	TIM	4.23
Pam	Female	42	-	-	-	-	-

Note:

* Names have been changed to maintain confidentiality

** Mode of administration: TBM = Tidal Breathing Mode; TIM = Target Inhalation Mode

3.2 Pen Portraits

The purpose of providing a pen portrait of each parent-adolescent dyad is to introduce the reader to the sample of participants by providing some more detailed background information. By including a description of some demographic details, background and interests and the parent and adolescent experiences of CF it is hoped that a more complete picture is provided. This section also includes some of the author's own impressions which were formed throughout and immediately after the semi-structured interview and written down within a reflective research journal. The participants' interactional style – including non-verbal aspects of the process – along with the richness and quality of their narrative account are reflected upon here. There will also be a summary of the emerging main themes that the analysis of each individual transcript uncovered in addition to some reflections on the parent and adolescent relationship.

Karen and Mary

Karen

Karen was aged 16 at the time of the interview and was studying at a local community college. She was diagnosed with CF shortly before her second birthday. She described not being affected particularly badly by CF but found the treatment regimen a burden and said that she goes through stages of not taking her treatment. Approximately three years ago she was diagnosed with CF related diabetes and this caused her significant emotional distress. She struggled to effectively manage the diabetes in the initial stages following diagnosis and was recently fitted with an insulin pump which she says has helped her to feel more in control of her treatment.

Karen described leading a very active social life. She was keen to be interviewed before her mother as she was due to go out and meet with her boyfriend and friends that evening. She had commented before the interview started that she really did not know why she often failed to take her aerosol therapy and was concerned that she would have little to say. However, as the interview progressed she was able to speak quite openly about CF and her treatment. Although at times she was unable to maintain eye contact and appeared a little distracted, she was not particularly difficult to engage in conversation and provided some detailed responses to the interview questions.

Karen did not like the fact that she had to sit and concentrate in order to effectively take her aerosol therapy. She described it being different to her tablets, inhaler and insulin

pump because it took longer to do and was inconvenient. She described often forgetting to take her aerosol therapy and when she did remember she would get distracted quite easily by the television, mobile phone or puppy which resulted in lengthier and less optimum treatment administration. On the one hand she said that she needed to be reminded to take her medication but on the other hand she found being prompted frustrating and at times this was a source of conflict with her mother.

Mary

Mary is a single mother of two and was aged 43 at the time of the interview. She lives with her eldest son (19) and Karen. Mary found it difficult to hide her frustration relating to her daughters non-adherence and at times got quite emotional in the interview. She was hopeful for Karen's future but was holding on to a great deal of worry and anxiety regarding the very serious potential health consequences of her not taking her treatment. My initial impressions were that she was exasperated by the daily process of taking responsibility for her daughter's treatment and the constant struggle to attempt to encourage her to adhere.

The analysis of the transcripts confirmed these initial impressions. Mary seemed to have got to a point where she recognised the need for her daughter to start taking greater responsibility for her treatment but was reluctant to relinquish control. She described the burden of guilt she carried when she knew that Karen had not been taking her aerosol therapy and this could sometimes lead to conflict with her daughter. Karen's recent request to take more responsibility of her treatment was welcomed on the one hand but not taken particularly seriously on the other. Karen's busy lifestyle and the impression she gives that taking her treatment is not an important priority seemed to be preventing her mother from trusting her with more responsibility. However, when tensions in the house were high, Mary said that she had begun to stand back and at times disengage from the treatment process as she feels that she has tried her best and failed.

Tom and Pippa

Tom

At age 11, Tom was the youngest participant to be interviewed. He had recently moved up from his Primary School and said he was enjoying the increased responsibility of High School. He described in detail his busy school days which he said started early because of his CF treatment. Although he was diagnosed with CF at age 7, he said he could not really

remember this and commented that living with CF was “a bit of a pain”. He said that his friends did not really understand his illness and throughout the interview it was apparent that he saw CF as making him different from others.

Tom really enjoyed playing on his Xbox and watching TV but also said he spent a lot of time playing outside and meeting up with his friends at the skatepark to ride on his scooter. He described resenting his treatments, particularly his physiotherapy and his nebuliser because they took up so much of his time. Although he said that his nebuliser was quick and easy to take, he did not like the fact that he had to take this alongside his physiotherapy. He described being in a consistent routine with his treatments but that this sometimes changed at the weekend depending on what activities he or his family was engaged with.

Overall, Tom was a pleasant character who spoke with maturity about his aerosol therapy treatment. He was uncertain as to the actual purpose of his aerosol therapy treatment but had a sense that it “kept him healthy”. Although he appeared a little under-confident at the beginning of the interview, he became much more engaged in the process as the interview progressed.

Pippa

Pippa lived with her two daughters (7 and 9), Tom and her husband. She was aged 39 at the time of the interview. She described a very busy daily routine which at times she said she felt overwhelmed with. Similarly to Tom, she described having some resentment towards his aerosol therapy treatment. This ill feeling was mainly directed at the amount of time it took to effectively clean the aerosol therapy device and the fact that she was unsure whether the treatment was actually having any beneficial effect. A number of recent hospital admissions for her son coupled with the burdensome nature of the daily treatment regimen appeared to be contributing to her current state of mind. However, it was clear that she would do whatever it took to attempt to keep her son healthy and described feelings of guilt even at the thought of not adhering because of her own frustrations.

Pippa was engaged throughout the interview and provided a rich and detailed account of her and her son’s experiences of living with CF. I got the sense that she really wanted to help me to understand the daily struggle of living with the condition and some of the associated worry and anxiety. I came away from the interview with a genuine sense of admiration for Tom, Pippa and their family.

Amanda and Carla

Amanda

Amanda was aged 14 at the time of the interview. She described enjoying school with her favourite subject being art. She was keen to show me some of her favourite drawings and engage in general conversation reflecting her warm and friendly approach throughout the interview. Although she had a busy social life within and outside of school, at times she came across as quite a shy girl who was very modest about her own abilities.

Amanda described finding her CF a “bit annoying” but said that she had never lived without it so had just got used to living with the condition. She said she found it frustrating that her family and friends did not really understand what it is like living with CF and having to take treatments, including her aerosol therapy. She was very computer literate and said she spent a lot of time on Facebook. She had developed a friendship on Facebook with another girl her age that had CF and she said that it was good to be in regular contact with someone who understood what it was like to live with the condition.

Amanda said she did not particularly like having to take her aerosol therapy as it got in the way of her doing other things and found it a particular burden when she was prescribed more than one aerosolised treatment. She said she often got distracted whilst taking her aerosol therapy by the computer, mobile phone or the TV and that this often frustrated her mum. However, she said she was reasonably consistent when it came to adhering to her aerosol therapy treatment. There was some uncertainty about whether she found it easier to take her aerosol therapy during the school week or at the weekend, but said that it was probably easier when she was at school because she was in a consistent routine.

Carla

Carla was aged 45 at the time of the interview and lived together with her husband and Amanda’s brother who was 17. She worked part-time and although she did not particularly enjoy her job, she was very grateful that they were flexible with her hours when it came to caring for Amanda. She enjoyed exercising and had to start the day very early to ensure she could incorporate an early morning work-out before beginning the process of helping Amanda with all of her treatments.

Carla was a very straight-talking, practical woman who discussed the importance of establishing a regular routine when it came to Amanda’s CF treatment. She said she felt she did everything she could to make the process of Amanda adhering to her treatments as simple

as possible and described finding it frustrating when her daughter procrastinated with taking her aerosol therapy or forgot to take it altogether. She said she often worried about Amanda's CF and was keen to monitor her daughter's adherence to aerosol therapy; often using frequent daily reminders in an attempt to provide some peace of mind.

Carla described her and her daughter being very similar. She said they had a good relationship and were very close. Although she would sometimes get frustrated with Amanda when it came to taking her aerosol therapy, she described having a lot of empathy for her daughter about having to take so many treatments. She was very engaged throughout the interview and provided some very helpful insights. However, it was clear that she was keen to press on with the discussion as she had a number of treatment-related tasks to complete that evening in addition to cooking the family meal perhaps reflecting the daily effort involved with caring for a child with CF.

Elizabeth and Greg

Elizabeth

Elizabeth was 13 at the time of the interview and attended the local comprehensive school. She was a friendly and intelligent girl who enjoyed sporting activities and led a very active life. She took a very practical approach towards her CF and seemed keen to dismiss any of the potentially emotional aspects of living with the disease. She was prescribed two aerosolised treatments per day which she said she usually took in the evening. She had been diagnosed with CF related diabetes approximately two years ago and although she said that this was an inconvenience, she again described taking a pragmatic approach to the management of the illness. Her parents separated approximately 6 years ago and she spent roughly an equal amount of time living between the two homes during the week. She kept one I-neb™ device at her father's house and one at her mother's.

Elizabeth described her school week as very planned and organised. She said that taking her I-neb™ had been part of her daily routine for a number of years and felt that this had enabled her to treat adhering to her aerosol therapy as a "part of life". Analysis revealed a strong sense of knowing what her treatments were for and feeling that her aerosol therapy kept her well. There were themes of her two aerosolised therapy treatments being dependent upon each other and the necessary planning that was needed in order to reduce the inconvenience that this caused. Her sense that her aerosol therapy was usually taken in the evenings was confirmed by the downloaded data. Analysis revealed a very busy morning

routine which she said prevented the I-neb™ from being taken at this time. Evenings were seen as more relaxing, but there was still a very clear structure in place which involved her adhering to her treatments.

Although Elizabeth took on most of the responsibility for preparing and cleaning the I-neb™, support from her family was a consistent theme. It was clear that she valued occasionally being prompted if her father had noticed that she may have forgotten to take her I-neb™. However, I got the sense that Elizabeth was something of a perfectionist and she did comment that she gets frustrated with herself if she forgets to take her aerosol therapy. Although she said that she prefers not to take her I-neb™ in front of her friends, she described a consistent and supportive friendship group who knew about her CF.

Greg

Greg was the only father taking part in the study and was aged 46 at the time of the interview. Elizabeth spent three to four nights a week living with her father and his partner in their home, with the rest of the week being spent living with her mother. My initial impressions were that he was a very welcoming man who had a great deal of involvement with his daughter's treatment regime and life in general. He also appeared to be a very practical man and it was perhaps unsurprising that he placed a great deal of value on routinised behaviour when taking into account his ex-forces background. Although this straightforward and practical approach perhaps led to his accounts being less in-depth than some of the other participating parents, he was a very articulate man and engaged well with the interview process.

The analysis revealed substantial positivity toward the I-neb™ device with Greg referring to it as “a great piece of kit”. There were strong beliefs about the value of the aerosol therapy and the way that it prevented pseudomonas and hospital admissions. So strong were these beliefs that he said that he would “fight hammer and nail” to ensure the hospital staff continued to prescribe it. The TIM mouthpiece was a welcomed addition to the I-neb™ and he was in no doubt that this had reduced his daughter's administration times and given her back some free time during the evening. He valued being able to see the downloaded data and was clearly very impressed both with his daughters 100% adherence rate over the preceding week and the short administration times. He said that this represented a typical week and said that it would be more uncommon if his daughter had missed her treatment.

There were strong themes around viewing aerosol therapy as a part of life and allowing his daughter the responsibility of being able to get on with it and “not make a fuss about it”. Although he felt that there were few issues with adherence, he did say that mornings were too busy to effectively take the I-neb™, preferring evenings when things were more relaxed. He also felt that school days were easier to take the I-neb™ than weekends because the consistency of the routine during the week allowed for effective planning. The same was true for term-time versus holidays and he added that it was much more difficult to effectively prepare and clean the I-neb™ when on holiday. However, he did say that efficient planning could usually overcome these difficulties perhaps representing his strong beliefs that his daughter’s aerosol therapy was an essential part of life that could not be compromised.

David and Lindsey

David

David had been admitted to hospital for a routine procedure at the time of the interview, so I agreed to meet with him on the ward as this was more convenient for the family. He was 12 at the time of the interview. He was very easy to engage in conversation although he sometimes found it difficult to stick to the topic under discussion. He would often provide very succinct answers to open questions that were relevant to the study. He was very passionate about his guitar playing and said that he enjoyed school.

David appeared to be a very confident and independent boy who took an impressive amount of responsibility for his treatments. He said that he frequently prepared and administered the aerosol therapy treatment himself and then cleaned the I-neb™ device afterwards. He said that he did not mind taking his aerosol therapy as it tasted quite nice. He understood the purpose of taking his I-neb™ and said that he would usually adhere to the treatment every day. He enjoyed playing outside with friends and said that the sooner he got his aerosol therapy “out of the way” the quicker he could go out to play.

Lindsey

Lindsey was 37 at the time of the interview. She lived with her husband, her daughter who was 11, and David. She worked within the NHS and was very knowledgeable about her son’s condition. Although she was not currently at work, she said that she enjoyed her job. She came across as very outgoing and seemed keen at the beginning of the interview to engage me in conversation about the role of a psychologist.

Lindsey said that she felt that David did not really have a problem with adhering to his aerosol therapy as he usually took it every day. She described feeling guilty if her son did not take his treatment and was aware that he did not adhere to his aerosol therapy on the weekend before the interview. She said that it had been difficult to prioritise his treatments over the weekend because they had family staying with them at the house. She described herself as a “control freak” and said that she reminded David numerous times throughout the day to remember to take his I-neb™. She said that it was often very difficult to manage the daily competing demands of family life alongside a complicated treatment regimen. She described this as often leading to feelings of frustration but was keen to point out that she did not view David’s treatments as burdensome as they were there to keep him well.

Lindsey was very complimentary about the design and functionality of the I-neb™ device and said that she much preferred this to the older types of nebuliser. She described finding it difficult when less experienced doctors did not seem to appreciate the daily struggle of trying to manage a complicated treatment regimen but said that she had a positive relationship with her son’s care team in general and in particular, the hospital consultants. This reflected my impression of her as someone who was very knowledgeable and capable, but who also appreciated empathy and support.

Simon and Pam

Simon

Simon was 13 at the time of the interview. He was very creative and was keen to show me one of his recent paintings after the interview had stopped. He said that he would like to become an art teacher when he was older. He enjoyed watching TV and playing computer games in his spare time. Although he liked the outdoors, he said that he was unable to participate in sporting activities because of his implanted ‘Portacath’ (portable catheter appliance) and the fact that he often got out of breath. He described being used to his CF treatment and said that in addition to his daily regimen he also had overnight gastronomy feeds and received intravenous antibiotics at home once every three to four months. He said that he preferred his tablets to his other treatments because they were the easiest and quickest to take. He was able to articulate his thoughts well and although he appeared a little nervous towards the beginning, he appeared to develop more confidence as the interview progressed.

Simon said that he did not really mind taking his aerosol therapy. He said he has noticed that his nebuliser administration times had reduced significantly since using the TIM

mouthpiece and that this was positive because he ‘got to do more stuff’. He had a good understanding of the purpose of his treatment and said that he was just beginning to take more responsibility for the preparation of aerosol therapy. He said that he did not take his aerosol therapy when he was at his dad’s house because his mum did not want him to forget to bring his nebuliser home with him. He said that he did not really mind missing his treatment because it gave him a night off and enabled him to ‘relax’.

Pam

Pam was aged 42 at the time of the interview. She separated from her husband approximately seven years ago and lived at home with her older son (17) and Simon. She was coming to the end her degree course and was thoroughly engaged in the process of writing up her dissertation. She was interested in the process of the Clinical Psychology Doctorate; asking questions and enjoying exchanging stories about the challenges of academic study. This seemed to put her at ease and she went on to explain the difficulties associated with caring for a child with CF alongside numerous other commitments.

Pam outlined a busy CF treatment regimen which took up a lot of her and Simon’s time together. There was a theme around encouraging Simon to become more independent both generally and in taking greater responsibility for his treatment. This became more evident during the process of analysis with consistent themes about Simon’s age and stage of development and his mum’s belief that it was important to gradually start to ‘push him’ to take on more responsibility. She did not feel that her son’s adherence to aerosol therapy was particularly problematic. She was actually of the belief that Simon’s adherence would be 100% nearly every week but for the fact that he did not take his aerosol therapy when he stayed over at his father’s house on a Saturday night. This was mainly because she did not want her son to forget to bring his aerosol therapy device back with him, but it still caused her some frustration as she felt that he was only able to get a break from his aerosol therapy treatment and therefore his CF, when he stayed with his father. Although she reflected that she felt ‘silly’ saying this, it was clear that this topic was emotionally charged.

Overall, Pam came across as a very supportive and caring mother who had a very good relationship with her son. She discussed numerous challenges in relation to Simon’s aerosol therapy treatment but seemed to overcome these very effectively perhaps reflecting her reported beliefs in the necessity for the treatment.

3.3 Parent group analysis

This chapter will begin by outlining the theoretical formulation of the parent group data, which consists of 9 core categories. The parent group data will be reported first as the accounts are richer and more detailed and provide helpful context to the child group analysis where the accounts are – perhaps unsurprisingly – less comprehensive. Each of the core categories will then be expanded upon by describing the associated subcategories. Quotes taken directly from the parent interviews will be used to further illustrate the subcategories.

3.3.1 Theoretical formulation

The detailed process of coding provided 436 open codes and 112 axial codes. These were then revised to produce 9 core categories, and 42 subcategories. The theoretical formulation comprises a number of interacting processes described by the parents and can be seen as a diagram in Figure 7. The formulation illustrates the process of their child adhering to aerosol therapy, as experienced by the six parents interviewed for this study. The 4 process areas which contain the 9 core categories are shown in Table 5.

Table 5: Arrangement of the core categories into a theoretical formulation

Core Category	Process Areas
	Parent's interaction with the aerosol therapy
Category 1: Regimen characteristics	
Category 2: Device characteristics	
	Parent's cognitive, affective and behavioural processes
Category 3: Beliefs about aerosol therapy	
Category 4: Emotional drivers	
Category 5: Strategies and behaviours	
	Parent's interaction with their child
Category 6: Parents experience of their child's adherence	
Category 7: Parent and adolescent relationship	
	The context of adherence
Category 8: Everyday life	
Category 9: Environmental influences	

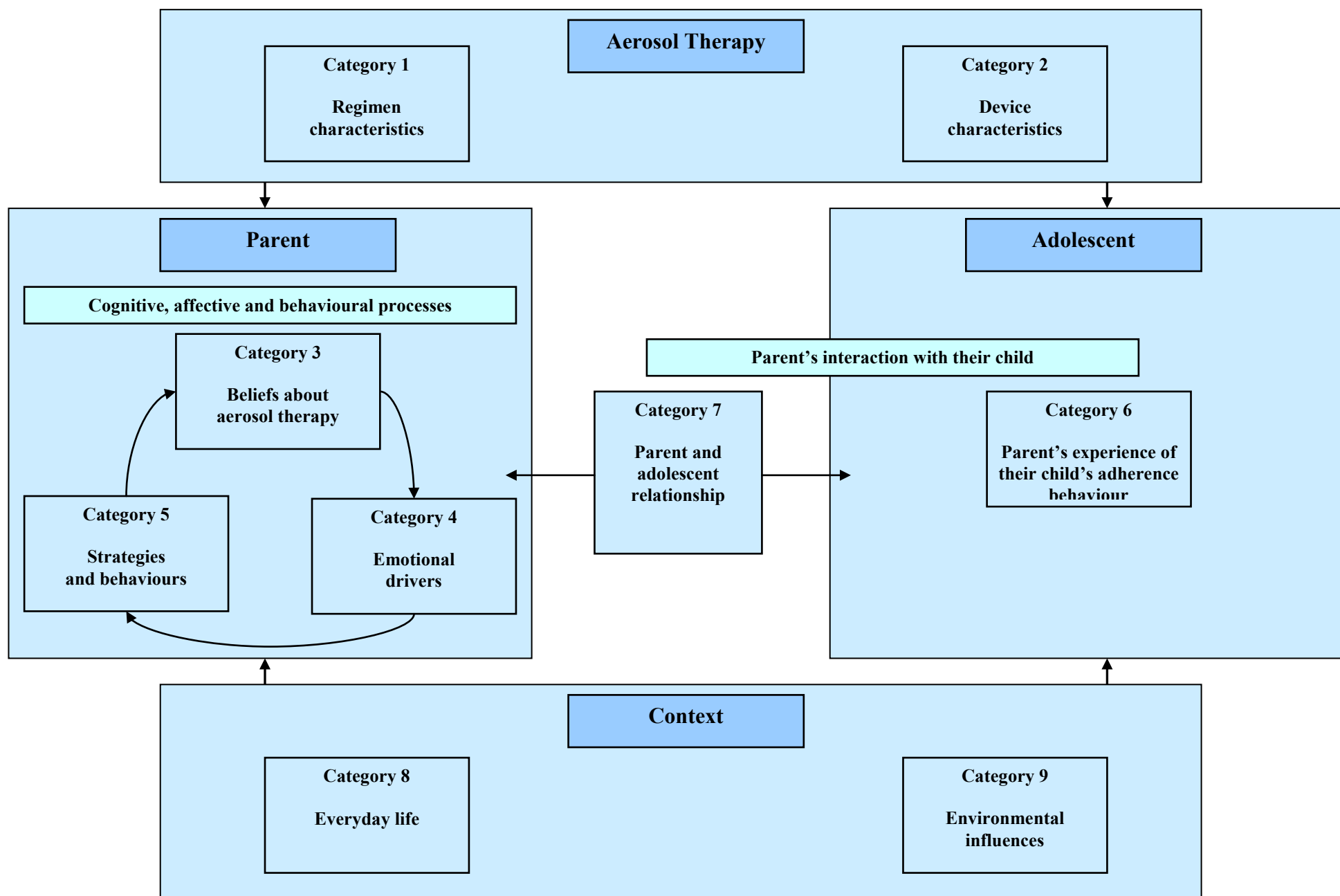


Figure 7: Overall theoretical formulation – Parent group data

3.3.1.1 Description of theoretical formulation

Parent's interaction with the aerosol therapy

Parents reported on the process of interacting with the aerosol therapy treatment regimen and the I-neb™ device itself. Many of the various characteristics of the regimen and some of the features of the device were seen as demanding and burdensome with parents reporting on the way in which these characteristics influenced their experiences, thoughts, feelings and behaviour around adherence. The experiences of the parents interacting with some of the more positive aspects of the I-neb™ device were described as helping to facilitate their child's adherence.

Parent's cognitive, affective and behavioural processes

The cognitive, affective and behavioural processes of the parents refers to the complex way in which their salient thoughts and beliefs about aerosol therapy interact with a number of strong feelings concerning their child's adherence to aerosol therapy. These interactions were seen as influencing a series of behaviours and strategies which had their relative strengths and weaknesses in facilitating the process of adherence. The linked, circular arrows within this section of the diagram represent the fact that these interactions are multifaceted and non-linear.

Parent's interaction with their child

This process area encompasses both the parent's experience of their child's adherence behaviour and the way in which the parent and adolescent relationship influences the overall process of adherence. Many of the experiences reported by the parents regarding their child adhering to aerosol therapy were a potential source of frustration. The arrows pointing outwards from category seven within the diagram represents the way in which the parent and adolescent relationship mediates the impact of the parent's experience of their child's adherence on their cognitive and behavioural processes and vice versa.

The context of adherence

Parents reported that the context in which adherence to aerosol therapy was taking place was important. Adherence did not exist in a vacuum and parents reported on the way in which everyday life and aspects of the environment interacted and influenced the process of adherence.

3.3.2 Summary

Overall, the accounts of the parents suggest a complex series of cognitive, emotional and behavioural processes which interact with the way in which they experience their child's adherence behaviour and are mediated largely by the relationship they have with their child. The above processes are further influenced by the interactions with the aerosol therapy regimen and device characteristics and the context in which adherence is taking place.

3.3.3 Core Categories

This section elaborates on each of the 9 core categories and 42 associated subcategories which have all been directly derived from the data. In order to demonstrate this, the number of parents who contributed to the categories is provided along with illustrative quotes taken directly from the transcripts. Whilst space prohibits the reporting of the full coding hierarchies – which illustrates diagrammatically the open and axial codes that contribute to each core category – an example of the full coding hierarchy used to generate core category 1 will be provided. In addition, the subcategories that contribute to the each of the remaining core categories will be also be represented diagrammatically.

Category 1 – Regimen characteristics

Number of sources: 6

Description: The experiences of the aerosol therapy treatment regimen were discussed by all of the participating parents. The parents reported on the multi-faceted nature of the aerosol therapy treatment regimen and these have been organised into six subcategories. The full coding hierarchy used to generate Core Category 1 can be seen in Figure 8.

Subcategory 1a: Multiple treatments

Number of sources: 6

Description: Parents reported that the aerosol therapy treatment was only one part of a complex and demanding CF treatment regimen that placed significant strains on the parent and adolescent:

“I think it’s very easy to look at things isolated, if you just had your 10 minute nebuliser every day or if you just had this tablet or if you just had that, but if you put the whole lot together it all gets too much sometimes because it’s all a big, it’s a whole, it’s just something on top of everything else.” (Carla)

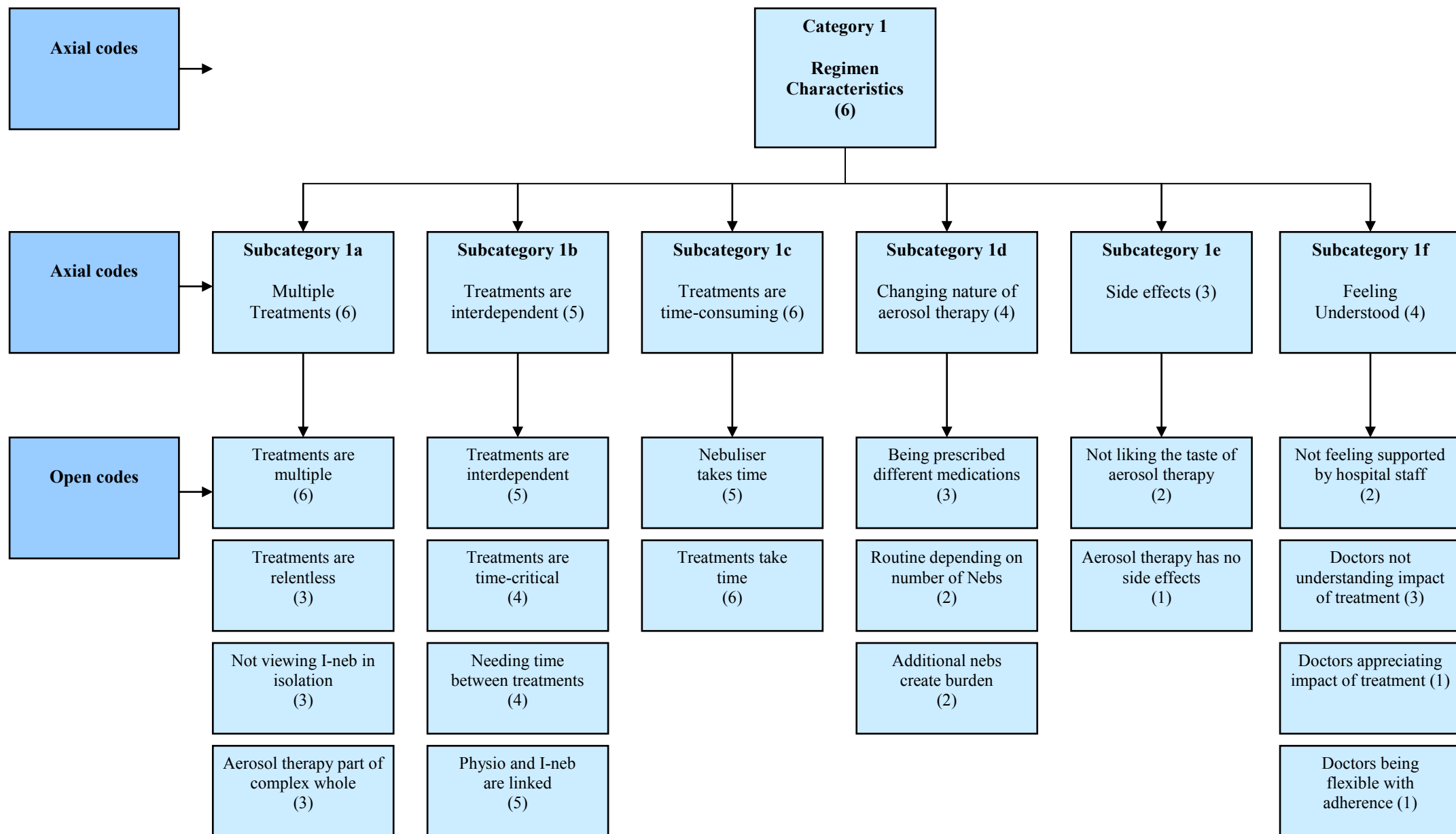


Figure 8: Coding hierarchy for Category 1 – “Regimen characteristics”

Subcategory 1b: Treatments are interdependent

Number of sources: 5

Description: Parents reported that the nature of certain treatments having to be administered in a time sensitive manner alongside other treatments placed further demands on the parent and adolescent:

“Erm, but it can’t be too close to his nebuliser either erm if you’re doing his physio it has to be at least an hour after his nebuliser.” (Pippa)

“As long as there’s a 2 hour period between his Colomycin and his DNase it’s ok.” (Pam)

Subcategory 1c: Treatments are time consuming

Number of sources: 6

Description: Parents reported that in general, the treatment regimen was time consuming with some parents specifically highlighting the time consuming nature of the aerosol therapy treatment:

“And I think, sometimes it, yeah you do resent how things are because it does take up so much time.” (Mary)

“I’d say it’s probably her physio and nebulisers that are the ones that she hates because they’re time consuming.” (Carla)

Subcategory 1d: The changing nature of the aerosol therapy regimen

Number of sources: 4

Description: Parents reported that the quantity and type of medication prescribed as part of the aerosol therapy treatment regimen often dictated the associated burden:

“When he first started with it I thought great. And then sometimes like when he’s had pseudomonas, he has three a day. He has like promixine twice a day and then he has his DNase. That’s pure evil. It’s like torture!” (Pippa)

“I say a lot of it depends on the drugs that they’re on and how time consuming erm and when they’re on you know two or three of them it’s all very well for [child’s name] at the minute because she’s only on one.” (Carla)

Subcategory 1e: Side effects

Number of sources: 3

Description: Some of the parents (2/3) reported that there were often side effects associated with certain nebulised treatments which negatively impacted on their child's experience of taking aerosol therapy:

"She was on tobramycin and that was horrible, it took such a long time to go through and apparently tasted horrible as well you know I take her word for it, I wasn't going to try it." **(Carla)**

This subcategory is also supported by statements made by one parent who did not feel that the aerosol therapy treatment had associated side effects:

"It's a treatment that they need to have you know, erm it doesn't take long, it's not painful, it's not time consuming. Erm there's no real reason why you wouldn't do it." **(Lindsey)**

Subcategory 1f: Feeling understood

Number of sources: 4

Description: Some of the parents (3/4) reported that the hospital doctors do not always seem to understand the impact that the aerosol therapy treatment regimen places upon parents and that this sometimes led to frustration and resentment:

"It is tough and I think when the doctors are saying you need to do this, this and this, they don't realise the day to day struggle of what it's really like to live with it. You know it's alright for them to sit there and say he needs this nebuliser every day, this tablet every day, that tablet every day, they're not dishing them up, administering all the medicine." **(Pippa)**

This subcategory is also supported by statements made by a parent who did feel that the doctors understood the impact of the aerosol therapy treatment regimen and allowed some flexibility with adherence:

"They've always said like for the matter of a week or two weeks if you're going abroad, it doesn't really matter about the nebuliser, you can afford to lose that period of time." **(Greg)**

Category 2 – Device Characteristics

Number of sources: 6

Description: All of the parents interviewed reported on their experiences of using the I-neb™ device. These positive and negative device characteristics were all experienced as important for the parents and have been organised into four subcategories (see Figure 9).

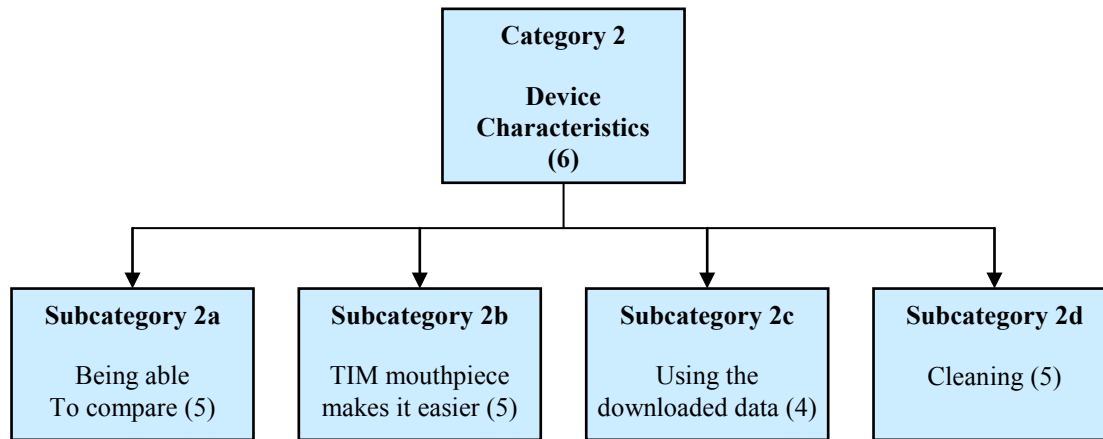


Figure 9: Subcategories contributing to parent data - Category 2

Subcategory 2a: Being able to compare technological advances

Number of sources: 5

Description: Parents reported that they were able to compare the older nebulisers to the newer I-neb™ device. They commented upon the technological advancement of the I-neb™ device and the associated benefits:

“The I-neb is, oh it’s so much easier than what it was because we used to have to set everything up and it’s mad, I can’t even remember how we used to set the old one up there was that many pieces erm and the I-neb is just fabulous.” (Pam)

Subcategory 2b: Target Inhalation Mode (TIM) mouthpiece makes it easier

Number of sources: 5

Description: Parents reported that the intelligent design of the TIM mouthpiece in providing users with immediate feedback on positive breathing technique had noticeably reduced aerosol therapy administration times:

“Since we’ve got the TIM, I’ve not had one argument out of him as to the fact that he’s got to do it.” (Pippa)

Subcategory 2c: Using the downloaded data

Number of sources: 4

Description: Parents reported that the data downloaded from the I-neb™ was interesting and helped to provide an objective measurement of adherence. This was the case both for the data used within the interviews and more generally with some (2/4) suggesting that it would be useful to discuss the data within clinic appointments more than it was currently:

“[Looking at the downloaded data] It’s really interesting actually because I just assume that she’s done it and she’s done it properly and she’s had the full dose erm but no that’s quite useful, it’s actually really interesting.” (Carla)

This subcategory is also supported by statements made by a parent who did not feel that the downloaded data was particularly helpful and viewed it in a more punitive way:

“The downloaded information? This is where Big Brother looks at you and points the big finger!” (Lindsey)

Subcategory 2d: Cleaning

Number of sources: 5

Description: Parents reported that cleaning the I-neb™ device thoroughly was of vital importance. However, the frequency and thoroughness of cleaning was experienced as burdensome:

“That’s another thing, you have to boil the damn thing once a week, you have to boil it for about 10, 15 minutes.” (Pippa)

Category 3 – Beliefs about aerosol therapy

Number of sources: 6

Description: All of the parents reported experiencing thoughts and beliefs specifically relating to the process of their child adhering to aerosol therapy. These salient thoughts and beliefs were experienced as part of a complex relationship between emotional and behavioural responses. This category has been organised into three subcategories (see Figure 10).

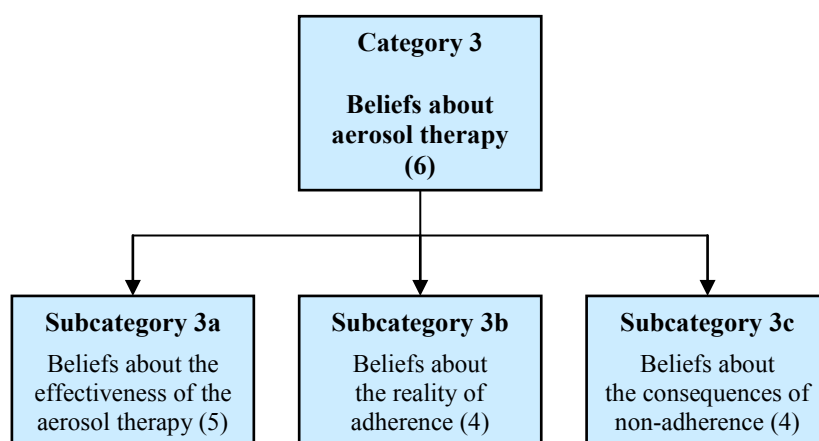


Figure 10: Subcategories contributing to parent data - Category 3

Subcategory 3a: Beliefs about the effectiveness of the aerosol therapy

Number of sources: 5

Description: Parents reported that their belief about the positive benefits gained from their child taking aerosol therapy was a factor which motivated them to encourage their child to adhere to the treatment:

“Anyway, as soon as they took her off the Neb, about a month after she was in hospital. Then they put her back on the Neb now ever since she’s been on the Neb, touch wood, right, she’s never been, she’s never had a stay in hospital so there’s got to be something in it as far as I’m concerned.” (Greg)

This subcategory is also supported by statements made by two parents who were still uncertain as to the benefit gained from their child taking aerosol therapy:

“I can understand why some parents would think ‘stuff it, it’s making no difference’ because in some ways I feel like that.” (Pippa)

Subcategory 3b: Beliefs about the reality of adherence

Number of sources: 4

Description: Parents reported that they thought it was unrealistic to expect 100% adherence to aerosol therapy every single day:

“But there are times when you can’t. You know, we’ve either been out for the day and it’s far too late or we’re all feeling under the weather, there are times where he doesn’t have it every day.” (Pippa)

Subcategory 3c: Beliefs about the consequences of non-adherence

Number of sources: 4

Description: Parents reported their thoughts and beliefs about the limited negative impact resulting from their child failing to adhere to their aerosol therapy treatment over a short period of time:

“You tend to think, ‘well you’ll be alright for a few days’ or whatever, ‘it’s only one treatment’.” (Lindsey)

This subcategory is also supported by statements made by a parent who believed that the ultimate consequence of her child failing to adhere to her treatment was a shortened life:

“The way she’s carrying on erm is very upsetting, ‘cos she won’t, she’ll be in her 20s.” (Mary)

Category 4 – Emotional drivers

Number of sources: 6

Description: All of the parents reported experiencing a number of emotions relating to the process of their child adhering to aerosol therapy and have been organised into 4

subcategories (see Figure 11). These emotions were often described in relation to the way they influenced certain behavioural patterns that the parents had noticed.

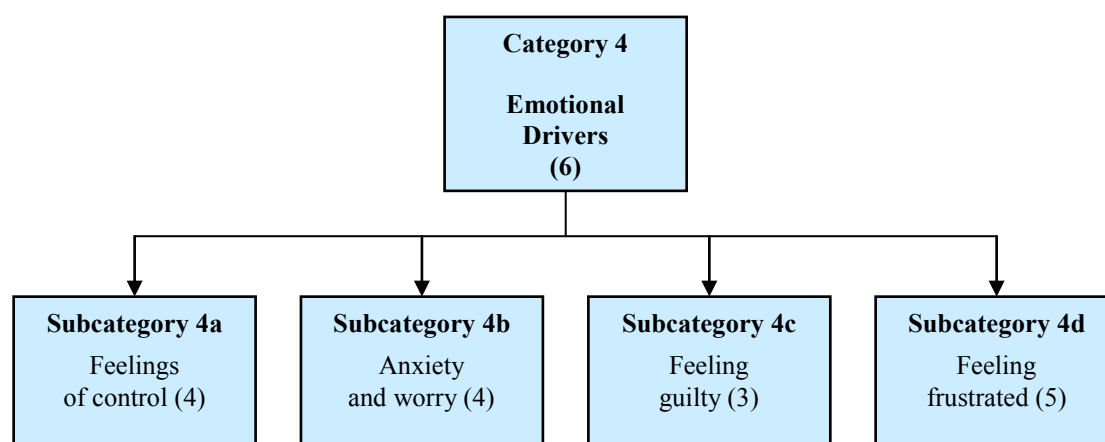


Figure 11: Subcategories contributing to parent data - Category 4

Subcategory 4a: Feelings of control

Number of sources: 4

Description: Parents reported that relinquishing the control they have over their child's adherence to aerosol therapy feels very difficult and often influenced a number of complex and interacting cognitive processes, emotional reactions and behavioural strategies:

"I like to know she's done it because if she hasn't done it I worry and again it's, she'll say I'm a control freak and I probably am a bit but having had all the years where I had complete control over her treatments to almost having no control at all now, erm yeah I like to remind her, just check in my own mind that she's done it."
(Carla)

Subcategory 4b: Anxiety and worry

Number of sources: 4

Description: Parents reported that at any given point, they were experiencing multiple worries relating to their child's CF. Parents also reported a number of worries specifically relating to their child's aerosol therapy:

"It's just something that's in the back of your mind all the time is 'what she's got in where she's going to be' and err you know and to 'make sure she's got it [i-neb] with her'." (Carla)

Subcategory 4c: Feeling guilty

Number of sources: 3

Description: Parents reported that they experienced significant feelings of guilt when their child had not adhered to their aerosol therapy, particularly if the parent had forgotten to prepare the medication or failed to remind their child:

“I feel really, really guilty if he doesn’t have it. Every now and again he might miss his nebuliser but I do feel really guilty when he does.” (Pippa)

“It makes me feel guilty when he hasn’t done his neb, that you’re failing him in a way.” (Lindsey)

Subcategory 4d: Feeling frustrated

Number of sources: 5

Description: Parents reported that there were numerous potential sources of frustration involved with the process of their child adhering to aerosol therapy. Some were related to the regimen and the device (see category 1 and 2), but many feelings of frustration stemmed from their child’s behaviour and relate to the later core category ‘Parents experience of their child’s adherence’ (Core category 6):

“[Looking at the downloaded data] I suppose she spent all morning in bed ‘cos she usually does as the weekend. In fact there’s absolutely no reason why she shouldn’t have done it.” (Carla)

Category 5 – Strategies and behaviours

Number of sources: 6

Description: The use of certain strategies in relation to the process of their child adhering to aerosol therapy was reported by all of the parents. Many of these strategies were interconnected to the thoughts and feelings outlined above with parents describing some of these strategies and behaviours as helpful and others as less helpful. In addition, there was some variation in the way that different parents experienced the same behaviours and strategies as either positive or negative in encouraging their child to adhere to their aerosol therapy. This category has been organised into seven subcategories (see Figure 12).

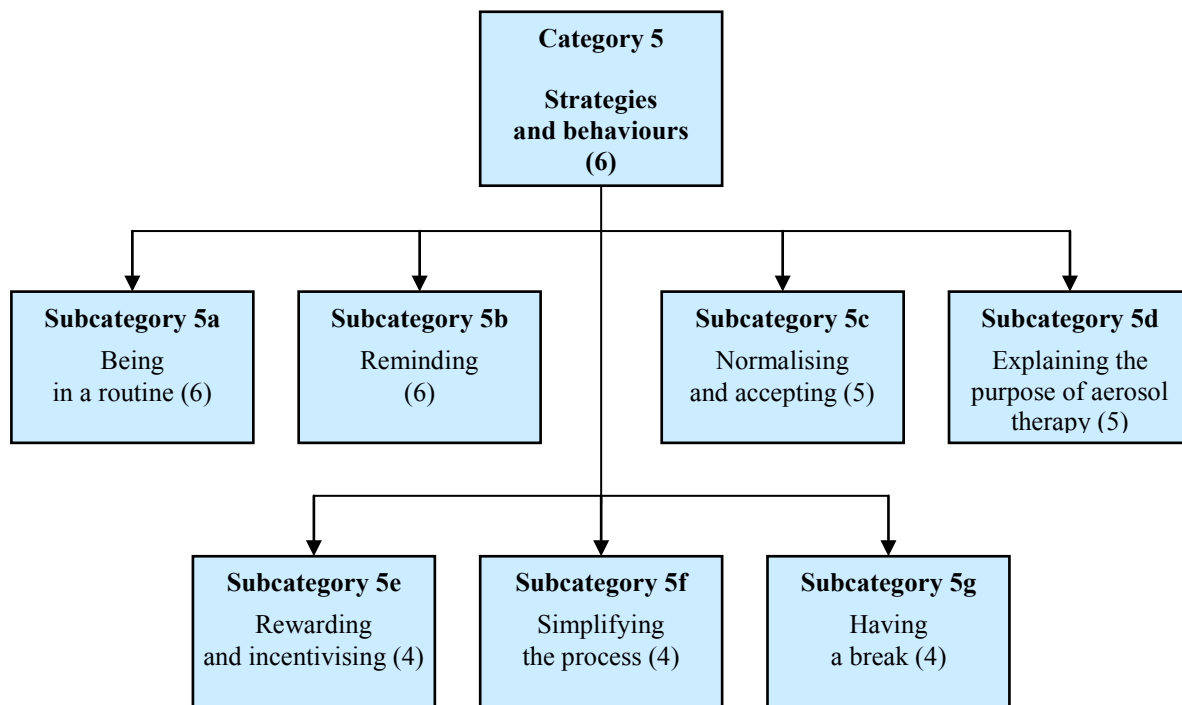


Figure 12: Subcategories contributing to parent data - Category 5

Subcategory 5a: Being in a routine

Number of sources: 6

Description: Parents reported on the importance of developing a well organised routine which helps to facilitate adherence. Although all of the parents discussed the importance of sticking to the established routine, some (2/6) also highlighted the necessity of allowing some flexibility within the routine to accommodate a busy lifestyle:

“You’ve got your routine of getting up, getting ready, going to school, coming home having your tea, having your Neb, having your physio, having your DNase, going to bed.” (Greg)

Subcategory 5b: Reminding

Number of sources: 6

Description: Parents reported that reminders were one of the main ways in which they monitored and supervised their child’s adherence. Most (4/6) parents suggested that they were a constant and repetitive feature of everyday life which sometimes led to conflict and did not necessarily encourage adherence:

“I should just record my voice and just play it continuously ‘[Name of child] do your blows, [Name of child] do your blows. Have you took your medicines? Have you done your neb?’” (Lindsey)

“She’s said if she doesn’t get reminded, she won’t take it but if she does get reminded, she resents being reminded to take it.” (Mary)

This subcategory is also supported by statements made by two of the parents who felt that the gentle and individualised way in which reminders were used and communicated to their child were a key factor in ensuring that this strategy encouraged adherence:

“You have to prompt her like and say ‘you should be doing something by now shouldn’t you?’. She’ll think ‘oh yeah’ and she’ll go and do it like you know.” (Greg)

Subcategory 5c: Normalising and accepting

Number of sources: 5

Description: Parents reported that trying to make the process of adhering to aerosol therapy as much a part of normal everyday life as possible was something that helped parents and their children accept the treatment and ‘get on with it’:

“I know it sounds peculiar but it’s just become part of normal life for us.” (Pam)

Subcategory 5d: Explaining the purpose of aerosol therapy to the child

Number of sources: 5

Description: Parents reported that it was of vital importance that their child understood the purpose of taking their aerosol therapy:

“If they’ve got no rationale about whether it’s beneficial to them, they just see it as an intrusion on their day to day life.” (Lindsey)

Subcategory 5e: Rewarding and incentivising adherence

Number of sources: 4

Description: Parents reported on the value of providing rewards and incentives for their child adhering to the aerosol therapy:

“[Looking at the downloaded data] She’d been told to get it done before dinner erm and she’ll have been doing it earlier here because she wanted to watch ‘I’m a celebrity’ and I told her to get it done.” (Carla)

Subcategory 5f: Simplifying the process

Number of sources: 4

Description: Parents reported that it was important to try to ensure that the process of adhering to aerosol therapy was made as simple as possible for their child. Some parents (2/4) reported feeling irritated if their child had failed to adhere despite them trying to make the process as easy as possible and relates to their earlier subcategory ‘Feeling frustrated’ (4d):

“I make it as easy as I can make it for her to do it.” (Mary)

Subcategory 5g: Having a break

Number of sources: 4

Description: Parents reported the mixed emotions involved with having a break from adhering to treatments. All of the parents said it was a relief to have a break but some (2/4) said that they experienced accompanying feelings of guilt as a result. In addition, one parent described feelings of resentment towards her ex-husband because her child did not adhere to his aerosol therapy when he stayed with him:

“I feel guilty that I’m putting the onus on somebody else to do his treatment when he’s not here but at the same time I’m really pleased that I don’t have to just be doing it every single day.” (Pippa)

“But I’d rather if he was, he could not have his treatment here as well rather than just at his dad’s. I can’t erm explain properly but erm it’s like he hasn’t got CF at his dad’s, he can have a night off from having CF and his treatments.” (Pam)

Category 6 – Parent’s experience of their child’s adherence behaviour

Number of sources: 6

Description: Parents reported on the numerous experiences encountered as a result of the process of their child taking aerosol therapy. Many of these experiences were a potential source of frustration for the parents. This category comprises six subcategories (see Figure 13).

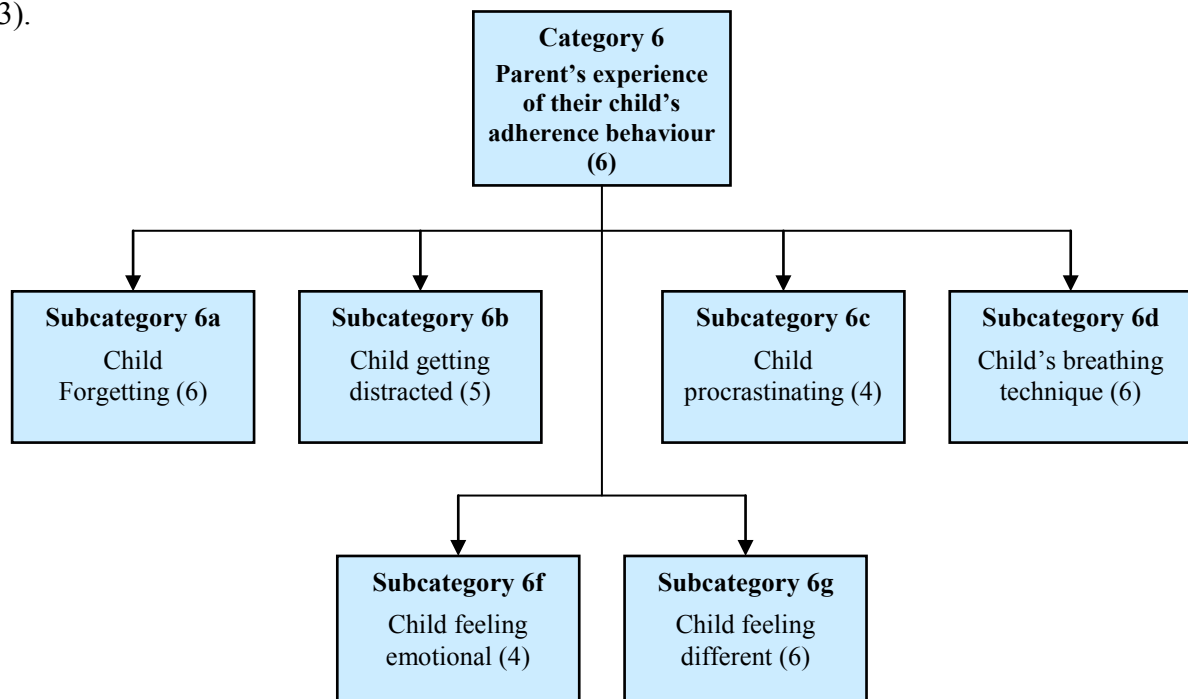


Figure 13: Subcategories contributing to parent data - Category 6

Subcategory 6a: Child forgetting

Number of sources: 6

Description: Parents reported that their child often forgot to take their aerosol therapy. Although parents reported that this was frustrating, there was an acknowledgment that this forgetting was often accidental:

“It gets forgotten sometimes. She doesn’t mean to forget it but you know she does forget it sometimes.” (Carla)

Subcategory 6b: Child getting distracted

Number of sources: 5

Description: Parents reported that their child often got distracted by various things and that this lack of concentration negatively impacted on adherence. This was often experienced as a source of frustration for the parents:

“She’ll be on her computer or something and then something comes up on Facebook and she’ll put it down for a second and she’ll ‘ooh yeah’ and pick it back up again.” (Carla)

Subcategory 6c: Child procrastinating

Number of sources: 4

Description: Parents reported that their child procrastinating about taking their aerosol therapy led to them feeling irritated:

“He would like prefer to sort of wait and wait and wait and wait and do it as late as possible. He’s trying to put it off.” (Pippa)

Subcategory 6d: Child’s breathing technique

Number of sources: 6

Description: Parents reported that their child’s poor breathing technique resulted in longer and less adequate aerosol therapy administration. Although parents experienced this as frustrating they described how the newer mouthpiece has improved their child’s breathing technique and relates to the earlier subcategory ‘TIM mouthpiece makes it easier’ (2b):

“He wasn’t trying hard enough to do really, really good breaths, it was taking a long, long time and a lot of the medicine was getting wasted.” (Pippa)

Subcategory 6e: Child feeling emotional

Number of sources: 4

Description: Parents reported that it was not uncommon for their child to feel upset and emotional. They reported that occasionally their child would get upset about actually having CF but more often they would become emotional about a whole variety of more normal, everyday concerns. Parents reported that their child's emotional state sometimes had an impact on the process of adhering to their treatments:

"When she's upset, she will not do any of her medication, her neb or anything. She won't take it." (Mary)

Subcategory 6f: Child feeling different

Number of sources: 6

Description: Parents reported that their child was often reluctant to take their aerosol therapy in front of others through fear of embarrassment. Parents also reported that taking their aerosol therapy in front of other people was a reminder to their child that they had CF and were different to other people:

"[Looking at the downloaded data] He wouldn't have taken it with him to his friend's house 'cos I think [Name of child] would be embarrassed if he ever took his nebs with him. Don't know if it's 'cos of his age but he doesn't like to talk to his friends about having CF." (Pam)

This subcategory is also supported by statements made by one parent who said that from her perspective, she did not think that their child was embarrassed about taking their treatments in front of other people:

"I don't think, he's never feels embarrassed about any of his treatments or anything like that." (Lindsey)

Category 7 – Parent and adolescent relationship

Number of sources: 6

Description: The complex and changing nature of the parent and adolescent relationship was reported by all of the parents. The relationship was seen as influencing and mediating the process of adhering to aerosol therapy. Perhaps unsurprisingly, aspects of the relationship that were categorised as constructive were reported to have a more beneficial impact on adherence than less positive aspects. This category comprises four subcategories (see Figure 14).

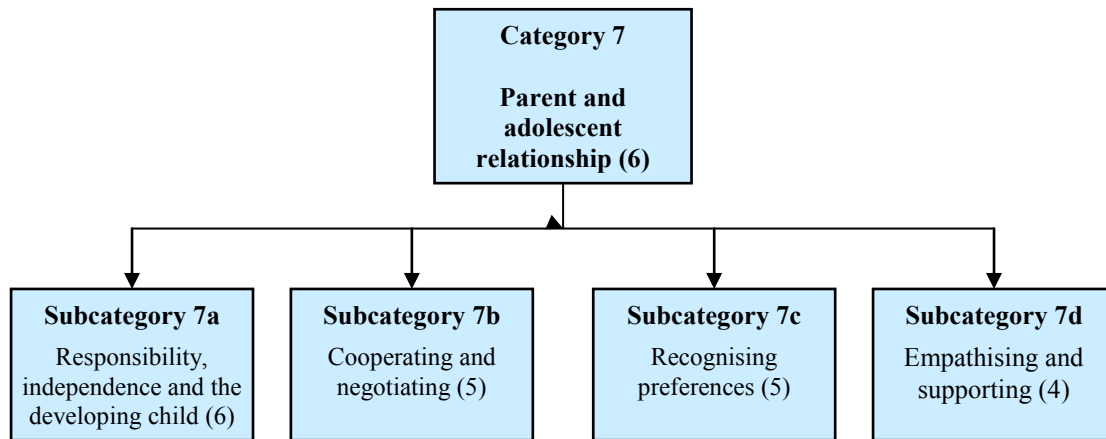


Figure 14: Subcategories contributing to parent data - Category 7

Subcategory 7a: Responsibility, independence and the developing child

Number of sources: 6

Description: Parents reported the importance of ensuring that increased responsibility around the aerosol therapy was gradually introduced as their child got older and developed greater independence:

"I think when they're younger, you have more control and that responsibility is then passed onto them. Like the I-neb, that's up to her to do it you know I can't make her do it and and whatever. The responsibility is on her now you know." **(Mary)**

This subcategory is also supported by statements made by many of the parents (4/6) who said that although introducing greater responsibility was important, it was difficult to do. This relates to the earlier subcategory 'Feelings of control' (4a):

"I think it's hard to let go, he's like nearly coming up to 13 and he's had his nebuliser from day 1 so it's hard not to have those erm those thoughts." **(Lindsey)**

Subcategory 7b: Cooperating and negotiating

Number of sources: 5

Description: Parents reported that it was necessary to cooperate effectively with their child around their aerosol therapy in order to avoid potential conflict and encourage and maintain adherence:

"[Looking at the downloaded data] I said 'while I'm in the shower, you make up your colomycin for me' so that's what he done." **(Pam)**

"But yeah the more you fight things the more she'll just dig her heels in. You've just got to make it as calm as possible erm" **(Carla)**

Subcategory 7c: Recognising preferences

Number of sources: 5

Description: Parents reported that it was important to recognise that their child had their own preferences concerning their CF treatment and more specifically had preferences when it came to the process of adhering to aerosol therapy. They reported that it was also important to acknowledge that these preferences may well be different to their own:

“I think his physio, he’s more inclined not to want to do that than his aerosol therapy because he knows that’s only going to take him a very short amount of time.” (Pippa)

“Whereas I would always do things straight away and get it out of the way, [Name of child] will always leave everything to the last minute.” (Carla)

Subcategory 7d: Empathising and supporting

Number of sources: 4

Description: Parents reported they could really empathise with their child about how hard it must be to adhere to a difficult treatment regimen and more specifically, adhering to the aerosol therapy. They reported that it was important to support their child as much as possible to help them deal with the daily struggle of treatment:

“I do tell [Name of child] as he does, what he has to go through ‘cos I am proud of him and he knows that erm and I do tell him so maybe that’s helped.” (Lindsey)

“I think that’s why I let him do it in front of the TV to try and make things a little bit more easier for him but I think it must be hard.” (Pam)

Category 8 – Everyday life

Number of sources: 6

Description: Parents reported on the way in which everyday life impacted on the process of adhering to the aerosol therapy treatment regimen and vice versa. This category has been organised into four subcategories (see Figure 15).

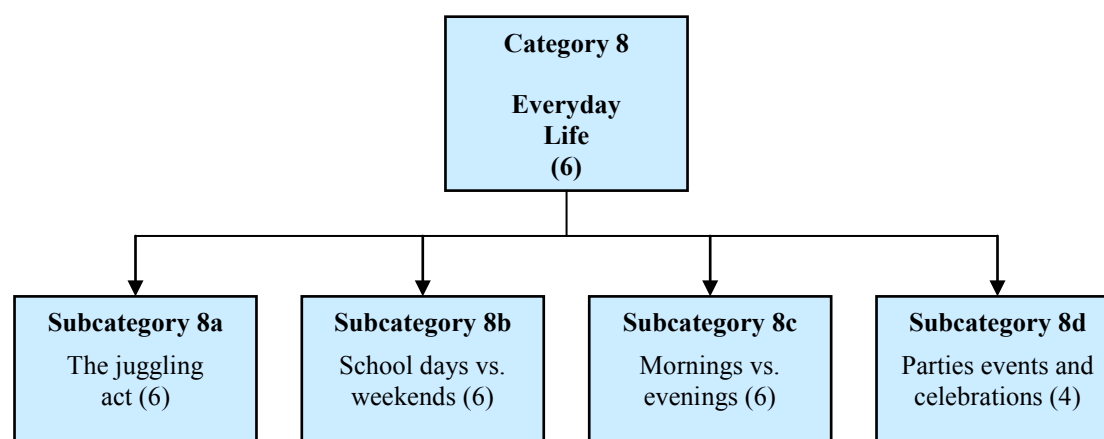


Figure 15: Subcategories contributing to parent data - Category 8

Subcategory 8a: The juggling act (fitting everything in)

Number of sources: 6

Description: Parents reported that trying to fit everything in during a typical day was similar to a juggling act which sometimes resulted in the child's aerosol therapy being dropped:

[Name of health professional] once asked us 'what gets in the way?' And we said 'life'. Life gets in the way because if you want to go out and do something or you want to go to a show or the cinemas, and you come back and it's 10 o'clock at night, you don't want to do your neb." **(Mary)**

This category was also supported by two parents who reported that despite having a busy lifestyle, it was important to try to ensure that the aerosol therapy treatment was incorporated into daily life and relates to the earlier subcategory 'Being in a routine' (5a):

"You go 'right ok, well how do we slot this into our lifestyle?' So you look at it then and you develop that routine and you stick to that routine." **(Greg)**

Subcategory 8b: School days vs. weekends

Number of sources: 6

Description: Parents reported that patterns of adhering to aerosol therapy were noticeable with half (3/6) suggesting that adhering during the school week was more straightforward:

"Weekdays are probably easier than weekends and school days are probably easier than when she's off school because you've got your routine haven't you? You've got your routine of getting up, getting ready, going to school, coming home having your tea, having your Neb, having your physio, having your Dornase, going to bed. Whereas on weekends, you're busy doing other things aren't you?" **(Greg)**

This subcategory is also supported by the other half of the parents who suggested that that weekend adherence was easier:

"I'd say the weekend when he's more chilled out 'cos school erm he does it again sitting on the couch but of a weekend he's more chilled and he'll just have his neb and he, I don't know if it's my perception but he seems to be more relaxed to take it of a weekend." **(Pam)**

Subcategory 8c: Mornings vs. evenings

Number of sources: 6

Description: Parents reported that there were further noticeable patterns of adhering to aerosol therapy with most (5/6) suggesting that taking the medication in the evening was

easier than in the morning. Mornings were thought to be too busy, whereas evenings were said to be more relaxed:

“[Looking at the downloaded data] We never do his Ineb in a morning. Because it’s just too much, when we’re getting three kids ready for school, ourselves ready for work.” (Pippa)

This subcategory is also supported by statements made by two parents who suggested that patterns of morning or evening adherence were also dependent on how many aerosolised therapies were prescribed and relates to the earlier subcategory ‘The changing nature of the aerosol therapy regimen’ (1d):

“If it was only once a day we’d always do it at night, if it’s twice a day obviously she’d have to do it in the morning as well.” (Carla)

However, this subcategory is also further supported by a parent who suggested that neither morning nor evening adherence was easier with both presenting difficulties:

“And we’ve tried, we’ve tried all the ways, we’ve tried doing it on a morning, we’ve tried doing it on a night, on a teatime when she came home from school. I don’t think there’s any good time for her.” (Mary)

Subcategory 8d: Parties, events and celebrations

Number of sources: 4

Description: Parents reported that special occasions often impacted on adherence to aerosol therapy with adherence often taking on less of a priority when there was an event or a celebration:

“Erm and it’s like when she goes to parties and things and when she’s going away with school and we try to let her drop as much as we can.” (Carla)

“[Looking at the downloaded data] That day was my birthday. I made [name of husband] do it [laughs]. ‘It’s my birthday, I’m not doing it, you can do it.’ (Pippa)

Category 9 – Environmental influences

Number of sources: 6

Description: All of the parents reported on the significant impact that certain aspects of the environment had on the process of adherence. The majority of accounts focussed on interactions with different and less familiar environments and the way in which they

influenced experiences of using the I-neb™ device. There are four subcategories that contribute to this core category (see Figure 16).

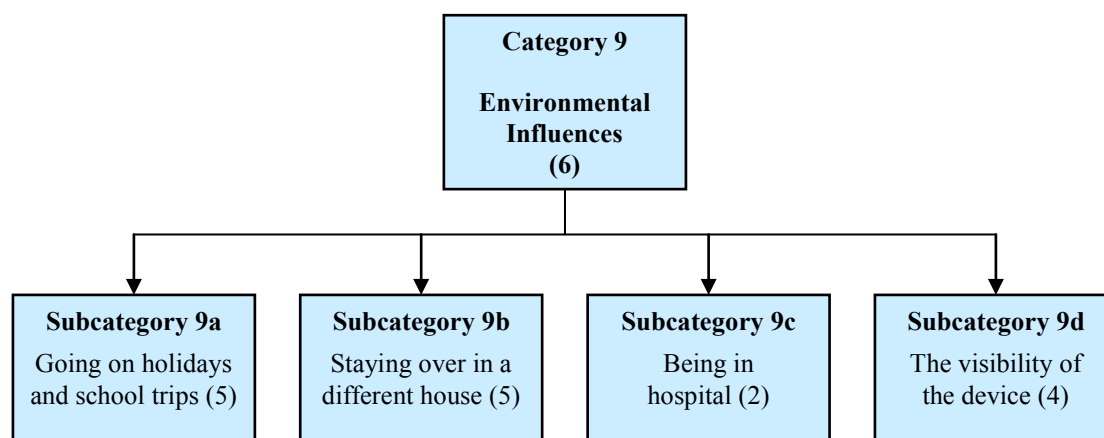


Figure 16: Subcategories contributing to parent data - Category 9

Subcategory 9a: Going on holidays and school trips

Number of sources: 5

Description: Parents reported that being away on holiday in an unfamiliar environment made it difficult for their child to adhere to the aerosol therapy because storing and preparing the medication and then cleaning the I-neb™ device was experienced as problematic:

“If we’re going away, if like we went camping, we didn’t take it with us, there’s nowhere to keep the medicine it’s got to be in a fridge. That’s a pain in the bum, because it’s got to be in a fridge all the time” (Pippa)

“When we’ve been on holiday or erm you know if she’s not had the facility for erm cleaning it, then obviously you might be only able to take it the once and then you can’t do it again because you can’t boil a kettle to you know clean the equipment properly and stuff.” (Greg)

Subcategory 9b: Staying over in a different house

Number of sources: 5

Description: Parents reported that staying over at someone else’s house made it more difficult for their child to adhere to their aerosol therapy. This included staying over at a friend’s house for a sleepover and also staying over at another parent’s house when the child’s parents were separated. Many of the reasons focussed again on the problem of storing, preparing and cleaning the I-neb™ device in an unfamiliar environment, but others included the child forgetting to bring the device back home and the child not wanting to take

the medication in front of their friends – which relates to the earlier subcategory ‘Child feeling different to others’ (6f):

“[Looking at the downloaded data] ‘Cos it looks like she hasn’t taken any of her treatments there. She was on a sleepover at her friend’s that night that would be the reason why.” (Mary)

“[Looking at the downloaded data] He goes to his dad’s on a Saturday night erm, I don’t let him take the Ineb with him ‘Cos he’ll forget to bring it back.” (Pam)

Subcategory 9c: Being in hospital

Number of sources: 2

Description: Parents reported that when their child was admitted to hospital, the lack of adequate facilities for preparing and cleaning the I-neb™ device made it difficult for them to adhere to the aerosol therapy:

“And interestingly, the worst time is when you’re in hospital with her and she’s obviously still using it but there’s no washing facilities there ‘cos you’ve got no plug that you can put into things.” (Carla)

Subcategory 9d: The visibility of the device

Number of sources: 4

Description: Parents reported that ensuring the I-neb™ device and associated equipment was visible in the home environment was something which helped to normalise aerosol therapy treatment but did not necessarily facilitate adherence:

“‘Cos it’s there, it’s in the kitchen, it’s there on view, I never put it away, it’s on view there and like when she has her supper, she must see it, but it just I just don’t think she just doesn’t think about it.” (Mary)

3.4 Adolescent group analysis

The analysis of the adolescent group data provided nine core categories. Each of the core categories will be expanded upon by describing the associated subcategories with quotes taken directly from the adolescent interviews being used to further illustrate these. Many of the themes within the adolescent group analysis were similar to the parent data – unsurprising given they are discussing their experiences of the same process – though the perspective expressed was often different.

3.4.1 Theoretical formulation

The process of coding provided 362 open codes and 87 axial codes. These were then revised to produce 9 core categories, and 37 subcategories. The theoretical formulation comprises a number of interacting processes described by the patients and can be seen as a diagram in Figure 17. This compliments the formulation described for the parent data and illustrates the process of adhering to aerosol therapy, as experienced by the six adolescent patients. Although there are differences at the level of content, the visual depiction of the theoretical formulation of the core categories is very similar to that presented on the parent data. Though some similarities were expected, the extent of the overlap was unexpected. This was led by the data, however, and represents the best fit for the reports provided by the adolescent participants. The 4 process areas which contain the 9 core categories are shown in Table 6.

Table 6: Arrangement of the core categories into a theoretical formulation

Core Category	Process Areas
Adolescent patient's interaction with the aerosol therapy	
Category 1: Regimen characteristics	
Category 2: Device characteristics	
Adolescent patient's cognitive, affective and behavioural processes	
Category 3: Beliefs about aerosol therapy	
Category 4: Emotional drivers	
Category 5: Strategies and behaviours	
Adolescent patient's interaction with their parent	
Category 6: Adolescent patient's experience of their parent's behaviour	
Category 7: Parent and adolescent relationship	
The context of adherence	
Category 8: Everyday life	
Category 9: Environmental influences	

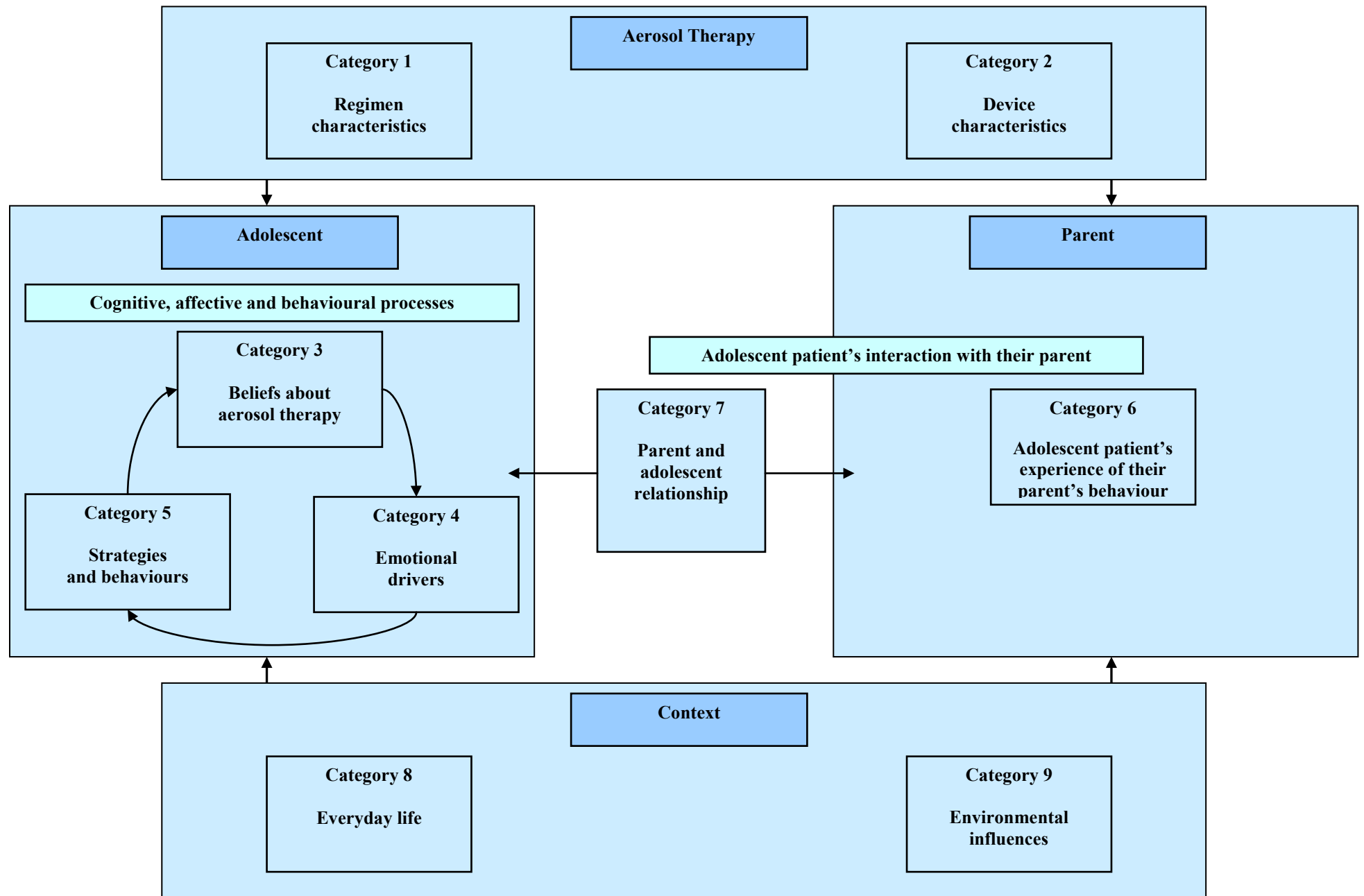


Figure 17: Overall theoretical formulation – Adolescent group data 82

3.4.1.1 Description of theoretical formulation

Adolescent patient's interaction with the aerosol therapy

Patients reported on the process of interacting with the aerosol therapy treatment regimen and the I-neb™ device itself. The accounts were similar to those of their parents with many of the regimen characteristics and some of the features of the device experienced as demanding. There was a particular emphasis on the lack of immediate feedback from the aerosol therapy medication. The feedback gained from the device itself was often experienced as positive. The way in which the above characteristics influenced the adolescents' experiences, thoughts, feelings and behaviour around adherence was very similar to the processes described by the parents.

Adolescent patients' cognitive, affective and behavioural processes

The cognitive, affective and behavioural processes of the adolescent patients were described in a less overt way than in the accounts of their parents. However, the data suggested that the complex way in which the patients' beliefs about adherence interacted with a series of emotional and behavioural responses was actually very similar to the processes described by the parents. The adolescents were obviously describing these from a patient perspective and this helped to provide valuable insight into the origins of some of the behaviours that negatively impacted upon adherence that their parents found confusing or frustrating. Again, the circular arrows within this section of the diagram represent the fact that these interactions are multifaceted and non-linear.

Adolescent patients' interaction with their parent

This process area is similar to the one outlined within the parent data section but this time describes the *adolescent patient's* experience of their *parent's* behaviour and the way in which the parent and adolescent relationship influences the overall process of adherence; this time discussed from a patient perspective. Interestingly, the reports concerning their parent's behaviour were often commented upon indirectly and those accounts that were more direct were largely positive. It is possible that this may have reflected the nature of the interview process and the adolescent patients' reluctance to be overly critical of their parents' behaviour to someone they had only just met. However, the accounts appeared to be very genuine and provided helpful insight from a patient perspective into the way in which the adolescent's interactions with their parent influenced the complex processes associated with adhering to aerosol therapy.

The context of adherence

Similarly to their parents, the adolescents reported that the context in which adherence to aerosol therapy was taking place was important. The way in which aspects of everyday life and features of the lived environment influenced the process of adherence was discussed from a patient perspective.

3.4.2 Summary

Overall, the accounts of the adolescents suggested that the interacting phenomena that were identified within the reports from the parents were also present as factors influencing adherence within the young patients. The adolescent patient accounts provided additional insight into the way in which the processes identified by the parents influence adherence to aerosol therapy.

3.4.3 Core Categories

This section elaborates on each of the 9 core categories and 37 associated subcategories which have all been directly derived from the data.

Category 1 – Regimen characteristics

Number of sources: 6

Description: The adolescent patients all reported on their experiences of the aerosol therapy treatment regimen. Although many of the regimen characteristics that were highlighted as impacting on the process of adhering to aerosol therapy were similar to their parents, there were some subtle differences in experience and perspective. The immediacy of the effect gained from taking the aerosol therapy was an additional area that the adolescent patients identified. This category has been organised into five subcategories (see Figure 18).

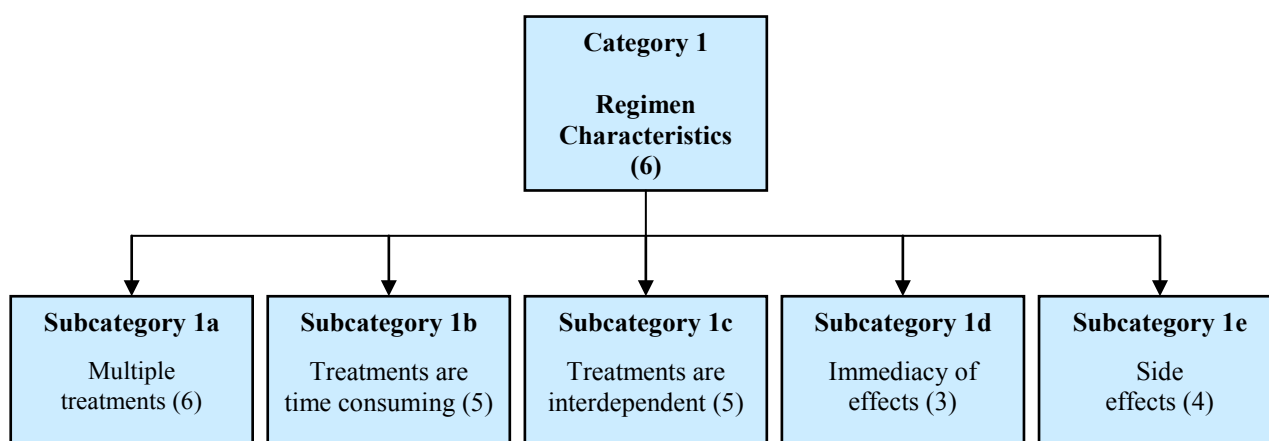


Figure 18: Subcategories contributing to adolescent data - Category 1

Subcategory 1a: Multiple treatments

Number of sources: 6

Description: Adolescent patients reported that they take numerous treatments for their CF. Similarly to the parents, many of the adolescents (4/6) reported that their aerosol therapy was just one further treatment to take as part of a burdensome daily regime:

“I have my IV’s about every three or four months which, it stays in for two weeks. Err, I have erm quite a lot of erm checkups, about six a year maybe. I have a nebuliser. Erm a physio with inhaler. I take about, probably about 10 normal tablets a day. Me enzymes, I probably have about 15 of them. There’s quite a lot really.”
(Simon)

“The nebuliser, it’s just another thing to do. It’s like, most people are just like ‘oh it’s just one thing, it just takes five minutes’ but it’s ANOTHER thing so.” **(Amanda)**

“Some days you get up and you have to do it but you just get bored of having to do them because you have to do so much like every day.” **(David)**

Subcategory 1b: Treatments are time consuming

Number of sources: 5

Description: Adolescent patients reported that the treatments taken to manage their CF are time consuming. Some (3/5) specifically highlighted the aerosol therapy treatment regimen as being more time consuming than other treatments, particularly if they were prescribed more than one nebulised medication:

“The nebuliser, you have to sit and concentrate and breathe for a while and it takes time. Like your tablets you can take them and they’re done and inhalers you can take them and they’re done but it’s a bit longer and inconvenient.” **(Karen)**

Subcategory 1c: Treatments are interdependent

Number of sources: 5

Description: Adolescent patients reported that some of their treatments are dependent upon each other; with physiotherapy and aerosol therapy being discussed in particular. Some (3/5) said that the process of taking their aerosol therapy is often influenced by their physiotherapy and vice versa:

“Erm it’s not the actual taking them it’s just time so if when I’m taking the first one which is normally promixin, I’ve got to leave an hour in between them and it’s got to be the last thing I do before I go to bed.” **(Elizabeth)**

“[Looking at the downloaded data] It depends on how long it’s taken me to do my blows. Like, if it takes me about 10 minutes or 15 minutes I end up doing my Neb about quarter past 7.” (David)

Subcategory 1d: Immediacy of effects

Number of sources: 3

Description: Adolescent patients reported that the lack of immediate benefits gained from taking aerosol therapy can negatively impact on adherence:

“Another thing is it, you don’t feel the immediate effects. Like an inhaler, more so with your inhaler, you just do it and you immediately, your lungs feel better but nebuliser it doesn’t, it’s over a long period of time you’d feel damage. It’s not like an immediate thing.” (Karen)

“The thing is it’s like erm Dornase it’s not an immediate dose kind of thing, it’s like a long term thing. I’ve been doing Dornase for so long so I figure I’ve built up enough to probably miss a couple of days.” (Amanda)

In contrast to the above reports, this subcategory is also supported by statements made by one patient who felt that the effects of his aerosol therapy were more immediate and that this encouraged him to adhere to the treatment:

“It can reduce the coughing quite a lot sometimes. I can be like quite chesty sometimes and after that I’m fine for a bit.” (Simon)

Subcategory 1e: Side effects

Number of sources: 4

Description: Adolescent patients reported that there were sometimes unpleasant side effects associated with taking their aerosol therapy:

“When I went into hospital we had to do this thing, this salty nebuliser. I didn’t like it ‘cos it made me cough loads.” (Tom)

“I always feel like it doesn’t take enough air into it as well, it feels like you’re suffocating a bit, I hate it.” (Amanda)

This subcategory was also supported by one patient who said that his prescribed aerosol therapy treatment had quite a pleasant taste which made it easier to take:

“I don’t mind it. I think it’s got quite a nice taste to be honest, Promixin. You can’t really describe it, it just tastes nice, it’s not a nasty taste. I don’t mind having to take that one.” (David)

Category 2 – Device Characteristics

Number of sources: 6

Description: Similarly to the parents, the adolescent patients identified a number of positive and negative characteristics of the I-neb™ device which they experienced as important in terms of influencing the process of adherence. The adolescent patient reports suggest a significant degree of importance is placed on the ability of the I-neb™ device to provide useful feedback. This category has been organised into five subcategories (see Figure 19).

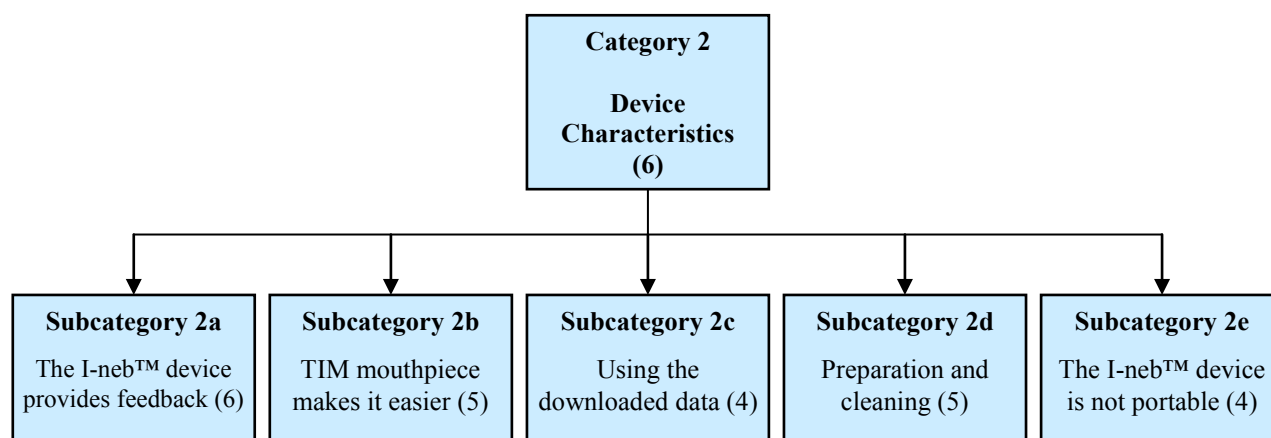


Figure 19: Subcategories contributing to adolescent data - Category 2

Subcategory 2a: The I-neb™ device provides feedback

Number of sources: 6

Description: Adolescent patients reported that the I-neb™ device was intelligent in the way that it provided feedback to help with accurate administration:

“Erm it buzzes and then I keep doing that until it like beeps once more and a face comes on the screen. It’s just a smiley face that tells you that you’ve done it.”

(David)

“Sometimes if I haven’t got the nebs in the right position it makes a little alarm.”

(Simon)

Subcategory 2b: Target Inhalation Mode (TIM) mouthpiece makes it easier

Number of sources: 5

Description: Adolescent patients reported very similar findings to their parents in terms of the way in which the TIM mouthpiece provided specific feedback around accurate breathing technique resulting in quicker administration times. This relates to the above subcategory ‘The I-neb™ device provides feedback’ (2a):

“We changed to this thing called the TIM mouthpiece. The new one shakes when you, err, when the breath has finished. So the old one took like five to ten minutes and the new one takes like half a minute to two minutes.” (Tom)

Subcategory 2c: Using the downloaded data

Number of sources: 5

Description: Adolescent patients reported mixed opinions about the data that is downloaded from the I-neb™ device both in terms of the current interview process and when this is done within their outpatient appointments. Most (4/5) said that they felt it was helpful as it provided a more accurate view of their adherence:

“[Looking at the downloaded data] I think it does help, like ‘cos you can see what its really like, I mean I’ve never thought of it like that. I never really looked at it like this before, just the times and like minutes that I’ve taken it in. 12 minutes. See when I’m on it, it doesn’t feel like 12 minutes.” (Elizabeth)

This subcategory also contains comments by two patients who felt that the downloaded data was not discussed within their outpatient appointments with one of them saying that it would probably not encourage adherence even if it was:

“I’m not sure why they always download it ‘cos they don’t really use it. I don’t know that it would make much difference.” (Amanda)

This subcategory is further supported by one patient who had mixed views about the value of seeing the downloaded information within her outpatient appointment but who went on to say that the approach taken by the health professional discussing it was important:

“I think I’ve done well and then you see it and I haven’t done as well as I thought. It does encourage me a little bit but it’s kind of disheartening. I don’t like going to the diabetic clinic at all, they’re always like ‘you can do better’. They never appreciate what I have done whereas [Name of health professional at the CF clinic] says ‘oh well, you’ve done it this many times but if you can, just try and do a bit better’. She doesn’t shout at you.” (Karen)

Subcategory 2d: Preparation and Cleaning

Number of sources: 6

Description: Adolescent patients reported that there was a necessary process of preparing the aerosol therapy medication and device in order for the treatment to be taken. Most (4/6) patients said that they were usually responsible for this preparation. There was an awareness

of the burdensome nature of cleaning the I-neb™ device but most (5/6) said that this was usually done by their parents:

“I put the medication in and take it and take it apart and then mum usually washes it.” (Amanda)

“I prepare it but I don’t wash it.” (Tom)

This subcategory is also supported by one child who said that they take responsibility for cleaning the I-neb™ device now that they are older:

“Yeah, I wash it. I never used to when I was little ‘cos I didn’t understand how to.” (Elizabeth)

Subcategory 2e: The I-neb™ device is not portable

Number of sources: 4

Description: Adolescent patients reported that the I-neb™ device was very rarely taken anywhere outside of their own house. Patients reported a number of device characteristics influencing this lack of portability including the necessity for storing all the associated aerosol therapy medication in the fridge and viewing the device as expensive and fragile:

“My tablets as well I can put them in my bag and take them with me. Or my inhaler I can take it with me but I don’t take my I-neb. I don’t tend to take it out, I usually leave it here. It’s because I’m a bit, ‘I’ll lose it!’. That’ll be my thing ‘cos it’s quite expensive, I wouldn’t want to lose it.” (Karen)

“I took it to a friend’s house erm but I don’t think I’d ever do it again because it was hard to keep track of where all my stuff was.” (Amanda)

Category 3 – Beliefs about aerosol therapy

Number of sources: 6

Description: All of the adolescent patients reported experiencing thoughts and beliefs that were specifically related to the process of adhering to their aerosol therapy. Although some of these thoughts and beliefs were discussed from a different perspective to their parents, the actual process of these beliefs interacting with a series of emotional and behavioural responses was very similar. In addition, the way in which the preferences for certain treatments influenced the process of adherence was also identified. This category has been organised into four subcategories (see Figure 20).

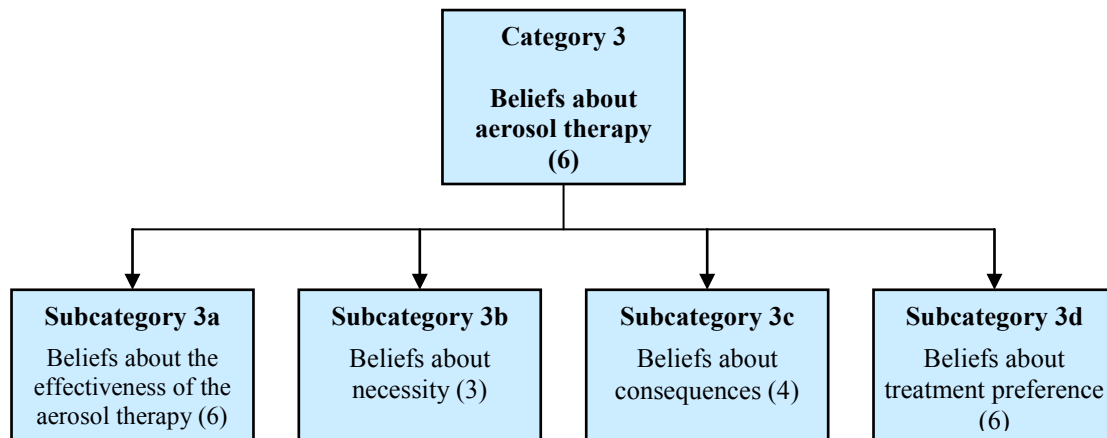


Figure 20: Subcategories contributing to adolescent data - Category 3

Subcategory 3a: Beliefs about the effectiveness of the aerosol therapy

Number of sources: 6

Description: Adolescent patients reported a number of beliefs about the effectiveness of the aerosol therapy. Most (4/6) believed that their aerosol therapy was important to take because it helped to keep them physically healthy:

“It’s really important because it obviously helps your health and looks after you. I haven’t been in hospital since I’ve been on them so.” (Elizabeth)

“I know it helps me quite a lot so I keep taking it.” (Simon)

This subcategory is also supported by statements made by two patients who reported mixed beliefs about their aerosol therapy, feeling that at times they were unsure as to the benefit gained from taking it. This relates to the earlier subcategory ‘Immediacy of effects’ (1d):

“I don’t feel like it does much but I know like when I don’t take it, I do notice that my chest feels a bit worse, but like for a day if I didn’t take it then I wouldn’t notice. I went through a phase of not taking it because I didn’t think it was doing anything.” (Karen)

Subcategory 3b: Beliefs about necessity

Number of sources: 3

Description: The adolescent patients reported specific beliefs concerning how necessary they felt it was to adhere to their aerosol therapy. Many of these beliefs related to the way in which they perceived their symptoms, often linking a greater necessity to adhere to aerosol therapy with feeling unwell:

“When I’m well it kind of slides a bit. It’s less of a priority, well it’s still a priority but it’s just not as important as when I’m unwell ‘cos when I’m unwell I want to get better but when I’m well it’s not as much of an incentive.” (Karen)

Subcategory 3c: Beliefs about consequences

Number of sources: 4

Description: The adolescent patients reported having specific thoughts and beliefs concerning the potential consequences of not adhering to their aerosol therapy treatment:

“I saw this programme once and it was about how things rot and I saw these erm like there were pseudomonas and that and erm it showed you how it looked through a microscope and stuff and proper bugs so ever since then like I’ve just been a bit more keen to do it.” (David)

This category was also supported by one of the adolescent patients who reported that even though she understood at a cognitive level the potential consequences associated with not adhering to her aerosol therapy it did not necessarily encourage adherence:

“’Cos like I’ve been told the consequences and stuff of not taking it and I still didn’t listen.” (Karen)

Subcategory 3d: Beliefs about treatment preference

Number of sources: 6

Description: The adolescent patients reported that they were influenced by a number of different internal beliefs about the preferences they had for certain treatments. Tablet medications, physiotherapy and aerosol therapy were the three main treatments that were often compared to each other with most (5/6) reporting that they preferred taking their tablets because these were more straightforward and took less time.

“To be honest, I probably out of all of them I’d rather have me tablets. They’re easier and they have less of an effect really. Like I can do them really quick whereas all me other ones I can’t really do much while I’m having them.” (Simon)

Many (4/6) reported that physiotherapy was the least favourite treatment because it was time consuming and often relied upon other people to help with the process.

“Doing me accapella, it takes me about an hour sometimes two hours. I just hate doing them, boring as well. So that’s pretty much the only treatment like that I don’t really like having.” (David)

Some (3/6) said that they did not necessarily dislike taking their aerosol therapy but only one reported having a preference for this over other treatments:

“I don’t mind having my Neb but it’s just the rest of them.” (David)

Two patients reported disliking their aerosol therapy in comparison to other treatments, citing as reasons the time taken to administer it and the fact that it made them feel different to others:

“I don’t mind taking my tablets or doing my insulin, it’s just that I wouldn’t sit there and do it [nebuliser] for five minutes when my mates are here.” (Karen)

Category 4 – Emotional drivers

Number of sources: 6

Description: The adolescent patients reported experiencing a number of emotions relating to the process of adhering to aerosol therapy. These emotions were quite different to those experienced by their parents. However, the way in which they seemed to influence certain behavioural patterns was very similar but appeared to be less conscious than the processes described by their parents. This category has been organised into four subcategories (see Figure 21).

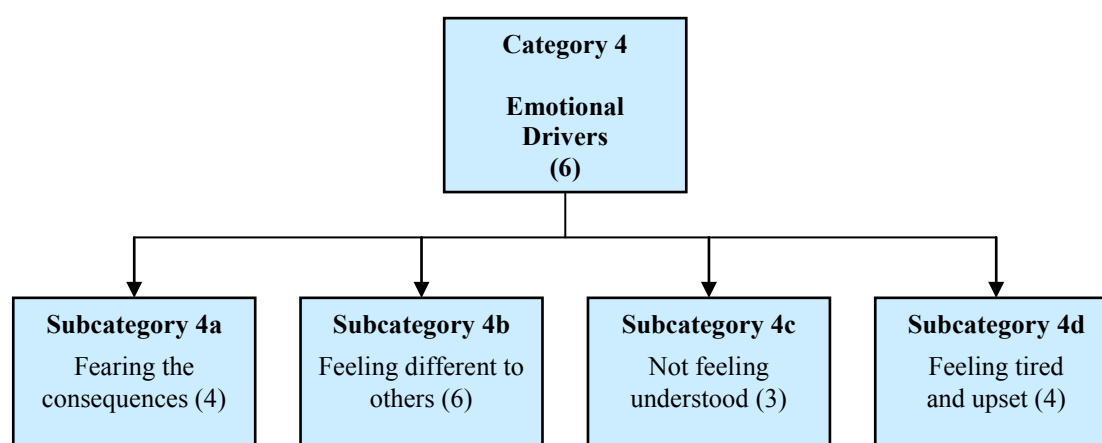


Figure 21: Subcategories contributing to adolescent data - Category 4

Subcategory 4a: Fearing the consequences

Number of sources: 4

Description: Adolescent patients reported that they were afraid of the numerous physical and social consequences associated with not taking their CF treatments and more specifically, not taking their aerosol therapy. Although this strongly relates to the earlier subcategory of

‘Beliefs about consequences’ (3c), the previous subcategory detailed a more *cognitive* engagement with thoughts and beliefs about the potential consequences of non-adherence whereas the current subcategory relates to the *emotional* fear concerning the potential consequences:

“At the end of the day if I don’t take them I could die because I would become ill and that but it’s keeping me alive but I don’t like doing them. If I don’t attack them [The pseudomonas] they’ll just erm kill part of my lung.” (David)

“I know that if I maybe stop taking it then I’ll grow pseudomonas again so it does encourage me to take them. I’m just frightened of missing school at the minute ‘cos it’s an important stage now in high school.” (Elizabeth)

Subcategory 4b: Feeling different to others

Number of sources: 6

Description: Adolescent patients reported that there were particular difficulties associated with taking their aerosol therapy in front of other people because it sometimes made them feel different and is related to the earlier subcategory ‘Beliefs about treatment preference’ (3d). Some of the patients (3/6) reported that aerosol therapy was harder to take in front of others than treatments in tablet form because it took longer. Two patients also talked about taking aerosol therapy in front of others, feeling that it was harder than physiotherapy because it was isolating, whereas certain types of physiotherapy could involve friends. Furthermore, some of the adolescent patients (2/6) reported that in addition to the difficulties associated with taking their aerosol therapy in front of friends, taking it in front of family members also singled them out.

“[Looking at the downloaded data] I’m allowed to do my Dornase during TV but I always feel quite bad ‘cos I just sit there and feel silly cos it buzzes, well not me buzzing, it just buzzes and it’s quite loud and dad just sits and looks at me.” (Amanda)

The language used by some of the participants emphasised the importance of keeping CF treatment separate from their social life:

“You could go on the trampoline and that, you could do that with a mate but you couldn’t take your nebuliser with a friend obviously.” (Tom)

Subcategory 4c: Not feeling understood

Number of sources: 3

Description: The adolescent patients reported that not feeling understood by family and friends was something that led to feelings of frustration and isolation, particularly when it concerned a lack of understanding about having to take treatments for CF:

“‘Cos it’s like, no one else understands really, it’s like my brother, he doesn’t understand at all, he’s just like ‘oh why can’t you just do this, it’s not a big problem’ but it’s like, adds up. And it’s like, like with my nebulisers it’s like I know mum thinks it isn’t much to do and I suppose it isn’t but it’s just another thing.” (Amanda)

Subcategory 4d: Feeling tired and upset

Number of sources: 4

Description: The adolescent patients reported that adhering to their CF treatments became even more difficult when they were feeling in a ‘bad mood’ or were feeling particularly tired:

“I just wouldn’t do them. It depends what mood I’m in really. Sometimes I don’t mind doing them but sometimes I do mind doing them. Erm sometimes when I’m tired, erm, or just don’t feel like doing them. I just get them over and done with quicker if I’m happy.” (David)

“That isn’t just a nebuliser thing, I do it with my tablets sometimes I just think, oh I’m too tired.” (Karen)

Category 5 – Strategies and behaviours

Number of sources: 6

Description: The adolescent patients reported engaging in a number of strategies and behaviours concerning the process of adhering to their aerosol therapy. Similarly to their parents, some of the reported strategies and behaviours were described as helping to facilitate adherence whereas others were describing as barriers to adherence. Perhaps unsurprisingly, they were less able than their parents to discuss in detail the ways in which their thoughts, beliefs and emotions about aerosol therapy influenced their behavioural patterns but they still provided some very useful insights. This category has been organised into five subcategories (see Figure 22).

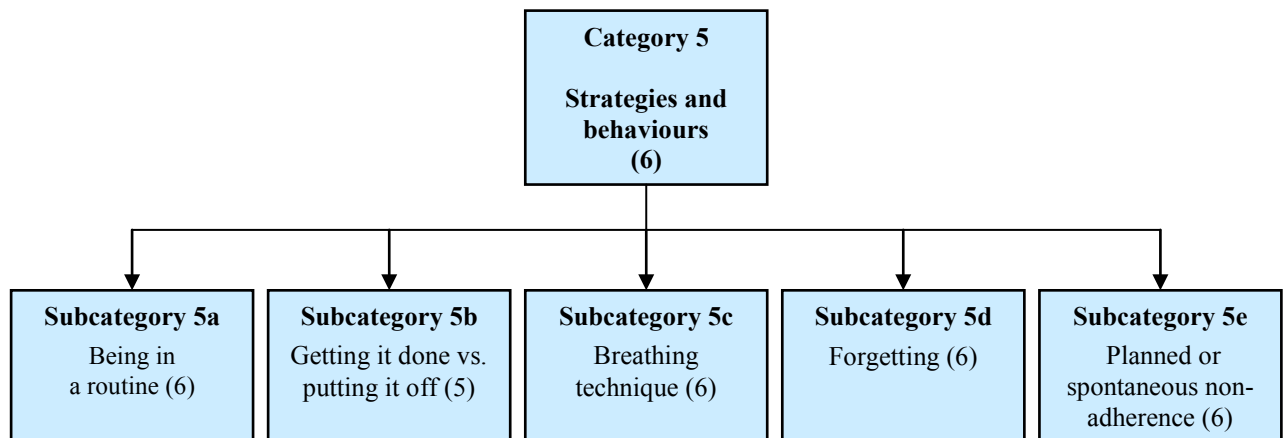


Figure 22: Subcategories contributing to adolescent data - Category 5

Subcategory 5a: Being in a routine

Number of sources: 6

Description: Adolescent patients reported that having a consistent routine helped to encourage adherence. Many (3/6) of the patients reported that there was more of an established routine with their treatments during weekday term-times:

“[Looking at the downloaded data] In the weekday I have much more of a routine I couldn’t describe my routine at the weekend, everything kind of just goes so I don’t know when I’m doing what, so I just try and fit it in when I can.” (Amanda)

“[Looking at the downloaded data] I take it, me first one which is just normally Promixin, I take after my dinner which is about half 5, quarter to 6 so sometimes I’ll be sitting here and it will hit 6 o’clock and I’ll be sitting here watching telly and I’ll go ‘Oh no’ so then I have to run into the kitchen and do it.” (Elizabeth)

Subcategory 5b: Getting it done vs. putting it off

Number of sources: 5

Description: The adolescent patients reported that they often engaged in two very separate strategies when it came to taking their aerosol therapy. Although some (3/5) appeared to have a preference for getting their aerosol therapy done quickly in order to free up their spare time, others (2/5) noticed a tendency to delay taking their aerosol therapy. While the data suggested that there were preferences, reports also indicated that these were not necessarily clear cut with adolescent patients sometimes alternating between the two strategies:

“‘Cos you can get it over and done with and you can do what you want then.” (Tom)

“[Looking at the downloaded data] I just leave it too late, that’s the only reason ‘cos I leave it too late and then I can’t be bothered so I just got to bed. But if I’ve got something to do I think ‘I’ll do it straight away’ and then I can go and do this.”
(Karen)

Subcategory 5c: Breathing technique

Number of sources: 6

Description: Adolescent patients reported that appropriate breathing technique was essential when using the I-neb™ device with all saying that it was important to take slow, deep breaths to ensure accurate administration. Although many (5/6) said that the TIM mouthpiece helped to facilitate an accurate breathing technique, some of these patients (3/6) also reported that trying to breathe quickly in order to speed up the process was still sometimes tempting:

“To make it easy you just have to breathe in like slowly instead of like fast because it like, it like it goes ‘HMMMMM’ and then it goes quiet and then it does a little shaking thing. So if you do that fast, then, it’s going to be harder. But if you do it slow then it gets to it.” **(Tom)**

“When I’m taking me Neb and I’ll be breathing, when I’m rushing I’ll be breathing faster to try and get it done and obviously that’s not the way to go about it because you’re not waiting for the buzz, so it takes longer.” **(Elizabeth)**

Subcategory 5d: Forgetting

Number of sources: 6

Description: Adolescent patients reported that they sometimes forgot to take their aerosol therapy. Although forgetting was largely accidental, two patients reported that forgetting to take their aerosol therapy also provided them with the opportunity to forget that they were ill:

“‘Cos I forget (laughs), I know it’s nice, not remembering I suppose. But it’s kind of hard to forget so I don’t really manage it. It’s nice to forget you’re ill, that’s good.”
(Amanda)

The above patient went on to further explain that the regularity involved with taking aerosol therapy over a long period of time actually serves to *facilitate* forgetting and that this causes her parents some confusion:

“That’s what mum and dad never understand they say ‘How can you forget? You’ve been doing it all your life’. It’s just I don’t know, maybe it’s ‘cos of that I just forget.” **(Amanda)**

Subcategory 5e: Planned vs. spontaneous non-adherence

Number of sources: 6

Description: The adolescent patients reported that there were occasions when they had either planned to not take their aerosol therapy or when the decision to not adhere had occurred more spontaneously. The reported explanations for spontaneous non-adherence were multiple and included being too busy, being late for something (e.g., catching the school bus, attending an out of school activity) and leaving it too late. Reasons for planned non-adherence included going away on holiday, staying over in a different house or having an event or celebration planned. Some (4/6) reported that having a break from adherence was welcomed. Three patients reported that planned non-adherence was often discussed more openly:

“Most people know about it. I think the people at the hospital know as well. They don’t seem too bothered that I have a night off. I quite like it because I can do a lot more. I can do what I want really.” (Simon)

Category 6 – Adolescent patients’ experience of their parent’s behaviour

Number of sources: 6

Description: The adolescent patients reported on their experiences of the behaviour of their parent; largely the behaviour of the mothers. As discussed earlier, many of the reports concerning their parent’s behaviour were commented upon indirectly and those accounts that were more direct were largely positive. This category comprises three subcategories (see Figure 23).

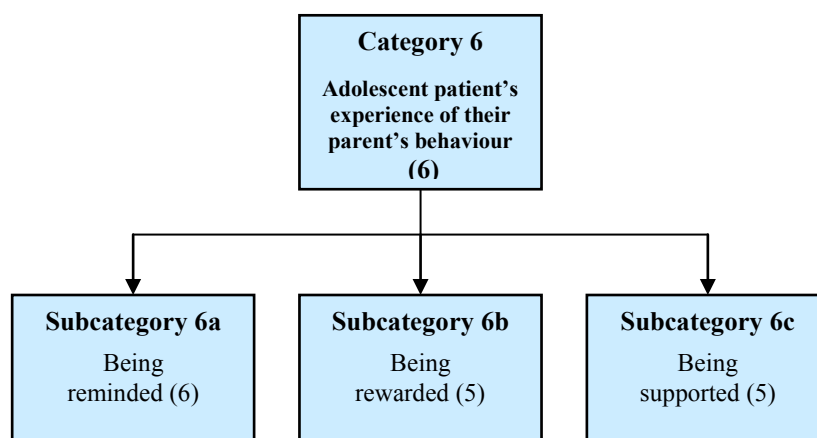


Figure 23: Subcategories contributing to adolescent data - Category 6

Subcategory 6a: Being reminded

Number of sources: 6

Description: Adolescent patients reported on the complicated relationship between reminders and adherence with most (5/6) saying that reminders were helpful:

“[Looking at the downloaded data] Erm on Sunday morning my mum reminded me, she said ‘Have you took your Neb?’, then I says ‘Oh God no!’. Erm, so I’ve gone and done it.” (David)

However, despite it being helpful this subcategory was also supported by statements from two adolescent patients who said that being reminded was still a source of frustration:

“I don’t like being reminded but I know that if I don’t get reminded then I won’t do it.” (Karen)

This subcategory is also supported by statements made by one of the adolescent patients who felt that the supportive way in which reminders were communicated helped to encourage adherence and was consistent with her father’s earlier account:

“Erm, my dad and [Name of Dad’s partner], they really help me a lot. They kind of, say if I’ve forgot and they say ‘Is there anything you need to do?’ and I immediately know.” (Elizabeth)

A statement made by one of the younger adolescent patients suggested that he is not afforded the luxury of being reminded to take his aerosol therapy treatment:

“I don’t get reminded, I just get told to do it.” (Tom)

Subcategory 6b: Being rewarded

Number of sources: 5

Description: The adolescent patients reported that their parents often rewarded and incentivised their adherence to aerosol therapy. This strategy was reported to have a motivating impact which encouraged adherence:

“Then after my dinner all my physio and I-neb and everything, after I’ve done all that, I can go back on my Xbox for like 10 or 20 minutes.” (Tom)

“[Looking at the downloaded data] I just like the kind of Saturday night TV stuff and my mum and dad don’t like me doing it [nebuliser] during TV so I have to get it done before TV.” (Amanda)

Subcategory 6c: Being Supported

Number of sources: 5

Description: The adolescent patients reported that their parents offered much needed support when it came to taking their CF treatments and more specifically, adhering to their aerosol therapy. The main types of support were practical (e.g., ordering the medication, cleaning the device) and emotional (e.g., empathising about treatment burden and providing comfort):

“I don’t know what I’d do without them ‘cos they’re a big part of it I suppose. They just help me in massive ways like sometimes I’ll feel a bit down about it and upset and they comfort me and stuff like that.” (Elizabeth)

Category 7 – Parent and adolescent relationship

Number of sources: 6

Description: The adolescent patients reported on their experiences of the relationship with their parents. Although many of the reports concerning the parent and adolescent relationship were again commented upon indirectly, the relationship was seen as influencing and mediating the process of adhering to aerosol therapy. This was very similar to the process reported by the parents with a more beneficial impact on adherence being associated with more positive aspects of the relationship. This category comprises four subcategories (see Figure 24).

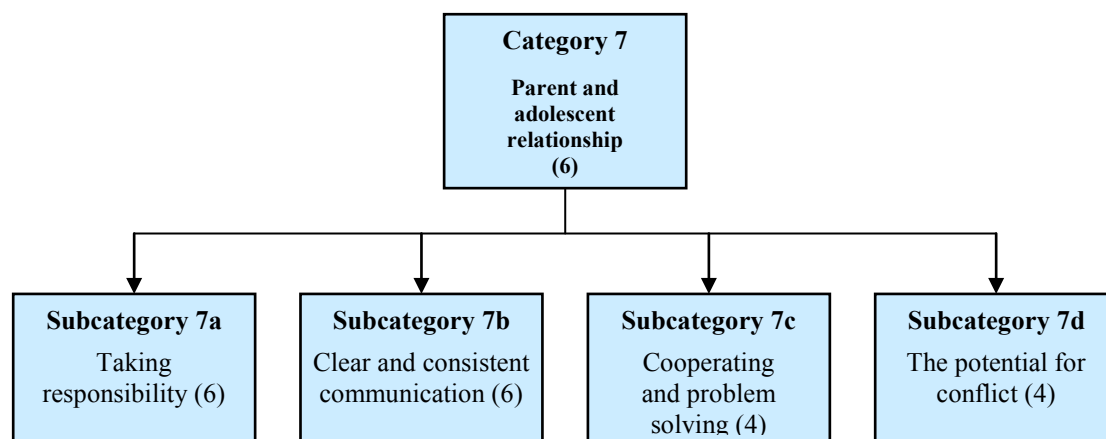


Figure 24: Subcategories contributing to adolescent data - Category 7

Subcategory 7a: Taking responsibility

Number of sources: 6

Description: The adolescent patient reports suggested that the parents – usually the mother – take the main responsibility for managing the process of adhering to aerosol therapy. This was particularly the case for those adolescent patients who were younger:

“I’ve just got to remember, but most of the time my mum’s in so I just do it when she gives it to me.” (Simon)

“If my mum probably wasn’t around then I probably wouldn’t do them.” (David)

The parent data reports suggested that relinquishing this responsibility and control as the child grew older was difficult. Some (2/6) adolescent patients reported that they recognised a need to take on greater responsibility as they got older:

“I’m going to have to learn to order it myself aren’t I? If I get into a routine of doing my neb every night it will help when I’m older won’t it?” (David)

This category is also supported by one adolescent patient who reported both recognising the need for taking on greater responsibility and who demonstrated an impressive ability to largely manage the process of adhering to the aerosol therapy herself. However, having the safety net of support offered by her parents was seen as reassuring and relates to the earlier subcategory ‘Being supported’ (6c):

“When I moved up to senior school and erm obviously I needed to start taking responsibility because my dad’s not always in the house so obviously I had to learn kind of how to do it.” (Elizabeth)

“I mean if I’m tired and I want to go to bed, sometimes they’ll wash my neb for me or make it up if I’m not well or whatever.” (Elizabeth)

Subcategory 7b: Clear and consistent communication

Number of sources: 6

Description: The adolescent patients reported that when it came to the process of adhering to aerosol therapy, having clear and consistent instructions and expectations from their parents was experienced as helpful. Patients also reported that they often became frustrated when this was not the case:

“She doesn’t, like, ‘cos if she were messing me around, like saying, ‘come and do your physio’, I do it. Then I go back up and then when I’ve just got back up to my room she says ‘come and do your nebuliser’, it would be quite annoying. But she says it as soon as I’ve finished my physio.” (Tom)

Subcategory 7c: Cooperating and problem solving

Number of sources: 4

Description: The adolescent patients reported that problems often got in the way of adhering to aerosol therapy. The successful negotiation of these problems was often achieved by the parent and adolescent working together to decide upon a solution:

“[Looking at the downloaded data] We had a conversation that we couldn’t leave too late because I hadn’t done my Neb and erm obviously the party was going to go ‘til like one, two in the morning. I think I just said that I hadn’t had my Neb and we said that we couldn’t leave too late then.” (Elizabeth)

Subcategory 7d: The potential for conflict

Number of sources: 4

Description: The adolescent patients reported that the process of adhering to aerosol therapy sometimes had the potential to cause conflict between the adolescent and their parent. The main sources of frustration for the adolescents came from parents ‘nagging’ or failing to recognise the burden of having to adhere to the aerosol therapy. The main frustrations for the parents were their child forgetting to take their aerosol therapy:

“My mum like nags me. Like mum says ‘take your neb’ and sometimes I like argue and say that I don’t want to.” (David)

Category 8 – Everyday life

Number of sources: 6

Description: Adolescent patients reported very similar findings to their parents in terms of the way in which everyday life impacted on the process of adhering to the aerosol therapy treatment regimen and vice versa. Patients reported an additional emphasis on the way in which aerosol therapy gets in the way of fun hobbies and activities. This category has been organised into four subcategories (see Figure 25).

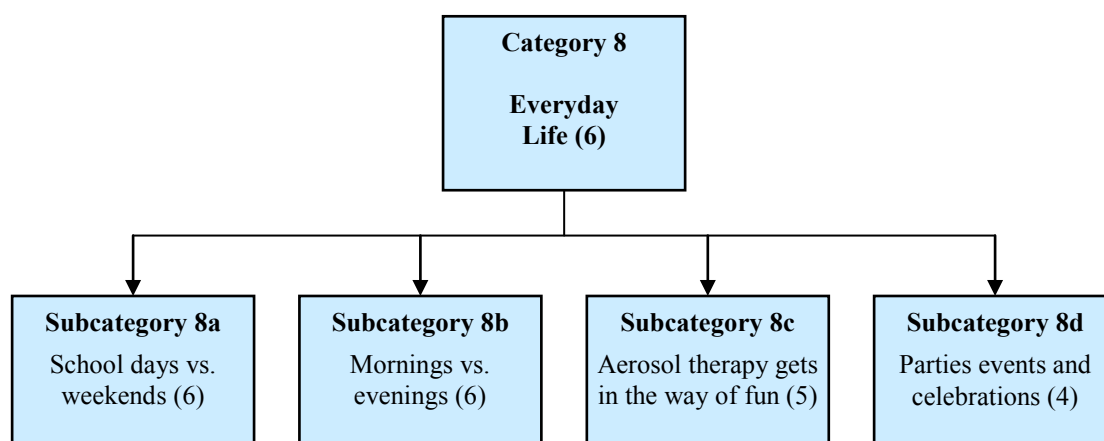


Figure 25: Subcategories contributing to adolescent data - Category 8

Subcategory 8a: School days vs. Weekends

Number of sources: 6

Description: Similarly to their parents, adolescent patients reported that there were differences in school day and weekend adherence. Some (3/6) reported that it was easier to adhere to their aerosol therapy during schooldays because of the consistent routine and relates to the earlier subcategory 'Being in a routine' (5a). When referring back to the parent data, this seemed to match their perceptions also but only for two out of the three patients:

"[Looking at the downloaded data] Erm I think it's better in school time because I've got a routine that's going on in school time." (Elizabeth)

In contrast, one patient reported that weekends were harder to adhere to because she felt she had less time. This was actually inconsistent with her parent's view:

"[Looking at the downloaded data] I think I find it harder at weekends 'cos we're strapped for time a bit at weekends." (Amanda)

This subcategory is also supported by some (2/6) patients who said that weekend adherence was easier and one patient who did not feel there were any differences between schooldays and weekends:

"I prefer doing it when I'm off school because I have more time." (David)

"I find them the same on weekends and weekdays." (Simon)

Subcategory 8b: Mornings vs. Evenings

Number of sources: 6

Description: Adolescent patients reported that there were times during the day that adhering to aerosol therapy was easier with some (3/6) suggesting that evening adherence was more straightforward:

"Probably in the evening 'cos I've got more energy whereas in the morning I've just woke up so I'm a bit drowsy." (Simon)

"I was like missing registration so I'd have to go and explain why I was late and things like that and so then we just decided to do it of a night time because it was easier and we had time on the night time." (Elizabeth)

Interestingly, this subcategory was also supported by statements made by half (3/6) of the adolescent patients who reported that they felt morning adherence was easier despite the downloaded data suggesting that they took their aerosol therapy in the evening. One patient

said that he realised he took it at night but just thought it would probably be easier in the morning whereas the other two were more surprised when presented with the downloaded information. This relates to the earlier subcategory 'Using the downloaded data' (2c):

"[Looking at the downloaded data] I don't know. It might be 'cos I lie in sometimes, when I don't have college. I have a lie in, so I don't get up 'til dinner time so then I'm a bit out of the routine. That could be why actually." **(Karen)**

Subcategory 8c: Aerosol therapy gets in the way of fun

Number of sources: 5

Description: Adolescent patients reported that having to take aerosol therapy is often experienced as inconvenient and can get in the way of enjoyable hobbies and activities:

"Like when I'm in the middle of something and I'm like doing really good on my Xbox or something. Like in a match and my Mum says to me to come down and do it and I say I'll just finish this match and I say it like 10 times and she keeps calling me down." **(Tom)**

"It's not particularly fun and it's just, I don't know it kind of gets in the way of me playing on the computer endlessly or erm like if I'm going to my friends." **(Amanda)**

This subcategory was also supported by two patients who said that although the I-neb can get in the way, the prospect of fun activities can also be used to motivate adherence and relates to the earlier subcategories 'Getting it done vs. putting it off' (5b) and 'Being rewarded' (6b):

"If I get it done quicker I can do more stuff like play me guitar, go on my Xbox, watch telly or play out like. If I get it done quick, I can just make the most of the day." **(David)**

Subcategory 8d: Parties, events and celebrations

Number of sources: 4

Description: The adolescent patients reported that celebrations and special occasions often impacted on adherence to aerosol therapy. Similarly to their parents, the adolescent patients reported that adherence took on less of a priority at these times and relates to the earlier subcategory 'Planned vs. spontaneous non-adherence' (5e):

"Going to the pub with my family or going to a party and being late or family coming erm when my family comes and stay over for the night I just want to spend time with family and forget about it." **(David)**

Category 9 – Environmental influences

Number of sources: 6

Description: All of the adolescent patients reported on the impact that certain aspects of the environment had on the process of adherence. Many of the patient accounts were similar to those reported by their parents, particularly in terms of the way in which less familiar environments acted as a barrier to adhering to their aerosol therapy. In addition, the adolescent patients highlighted the negative impact that easy access to technological devices within their environment had on the process of adherence. There are three subcategories that contribute to this core category (see Figure 26).

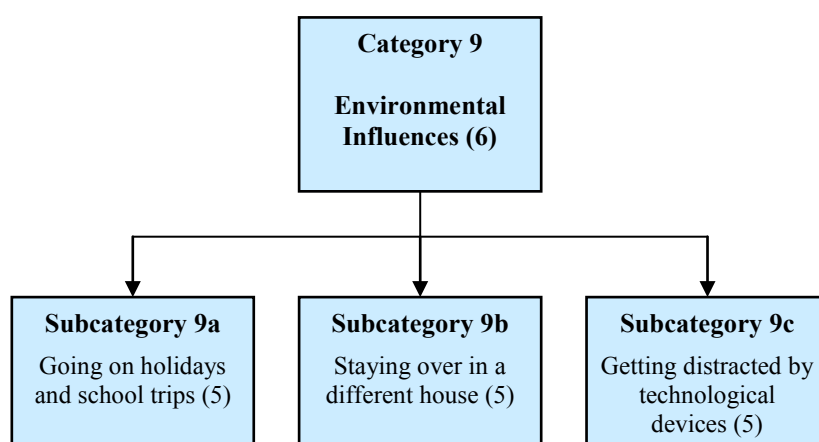


Figure 26: Subcategories contributing to adolescent data - Category 9

Subcategory 9a: Going on holidays and school trips

Number of sources: 5

Description: Adolescent patients reported that being away on holiday or going on a school trip made it more difficult to adhere to their aerosol therapy and was consistent with the views reported within the parent data:

“If I’m staying in a tent, maybe not take them for the weekend because it would be really hard to clean because obviously you’ve not any boiled water or any facilities like that.” (Amanda).

This category was also supported by one adolescent patient who reported that he did not experience any difficulties with adhering to his aerosol therapy when he was away:

“Erm when I’m on holiday I still do it the exact same really. Yeah, I go back to the room at night and have it really quick.” (Simon)

Subcategory 9b: Staying over in a different house

Number of sources: 5

Description: The adolescent patients reported that it was more difficult to adhere to their aerosol therapy when they were staying over at someone else's house. There were a number of reasons for this with explanations relating to the earlier subcategories 'I-neb™ device is not portable' (2e), 'Feeling different to others' (4b) and 'Planned vs. spontaneous non-adherence' (5e):

"[Looking at the downloaded data] I don't normally take me neb to me dad's err I think like it's like a bit of a night off it. Saturday I was staying at me friend's house. If I'm staying at me mates I don't really bring it either. I think that's also a bit of a night off I think sometimes as well. It's just relaxing really and I can just talk to me mates and that." (Simon)

Subcategory 9c: Getting distracted by technological devices

Number of sources: 5

Description: Adolescent patients reported that they would frequently get distracted when it came to taking their aerosol therapy. Becoming distracted was often reported by the adolescent patients in terms of them being passively influenced by aspects of their environment as opposed to them actively engaging in distractions as some kind conscious behaviour. Technological distractions (e.g., television, social networking sites and mobile phones) were the main sources with patients reporting that these things were often responsible both for forgetting to take the aerosol therapy altogether and for partial administration:

"[Looking at the downloaded data] I came back down to watch telly and [Name of boyfriend] said 'Do your I-neb'. Then someone could have rang, it does that a lot, someone rings me and I put it down. It could have been my dad that rang me'. (Karen)

"[Looking at the downloaded data] I was probably on my computer, I might have been texting someone. If I'm on Facebook and I see something interesting, I'll put it down to type something or if I'm like on a game I'll put it down to do something quickly and I'll forget about it." (Amanda)

4.0 DISCUSSION

The aim of the discussion chapter is to relate the findings of the current study to the existing literature. This chapter will begin by revisiting the research aim before providing a summary of the results. The main findings of the study will then be discussed in terms of the contribution they make to the existing knowledge base before moving on to summarise a number of methodological considerations, clinical implications and suggestions for future research.

4.1 Revisiting the research aim

The primary research aim of the current study was to qualitatively explore how the process of adhering to aerosol therapy in CF is experienced by both an adolescent patient and their parent. Although there were secondary research questions (e.g., the differences and commonalities in the experience of aerosol therapy compared to other therapies used in CF; and whether the use of the data downloaded from the I-neb™ device helped to qualitatively explore the process of adhering to aerosol therapy) the main focus of the discussion will centre around the primary research aim.

12 participants (six parent-adolescent dyads) were interviewed about their experiences of adhering to aerosol therapy. A seven-day retrospective download of data from the I-neb™ device was used to help guide part of the semi-structured interview. The Grounded Theory Method was used to analyse the interview transcripts and a theoretical formulation describing the participants' experiences of the process of adhering to aerosol therapy was developed. It is hoped that this will contribute to the wider literature concerning adherence to treatment in adolescents with CF.

4.2 Summary of results

The analysis revealed a complex interaction of cognitive, emotional and behavioural processes around adherence, with the parent and adolescent experience of one another at the core, and the parent and child relationship mediating the interactions. The above processes are further influenced by the interactions with the aerosol therapy treatment regimen, device characteristics and context in which adherence is taking place.

Participants were cued by the data downloaded from the I-neb™ device which helped to offer a window into their lived experience and enabled rich and open accounts of the numerous processes that influence adherence to aerosol therapy treatment to be gained.

Parents and adolescents often compared aerosol therapy to other forms of treatment, with an internal hierarchy of preference often favouring those treatments that were quick and less intrusive. Although the overall theoretical formulation was very similar for adolescent and parent, the analysis highlighted the way in which the same phenomena could be experienced from different viewpoints and provided valuable insight into the process of adherence from both perspectives.

The current study supports the view of Dziuban et al (2010) who suggested that treatment adherence is likely to be influenced by deep and complex psychosocial processes that are often difficult to ascertain. There is also support for the developmental, biopsychosocial model proposed by Barker & Quittner (2010), with the current study identifying a series of interacting biological (i.e., aerosol therapy regimen characteristics which are often dependent on disease management and progression), psychological (i.e., cognitive, emotional and behavioural processes alongside specific developmental challenges in the context of the parent-child relationship) and social influences (i.e., the environment in which adherence takes place and the demands of everyday life) on adherence to aerosol therapy treatment.

4.3 The aerosol therapy

Parents and adolescents reported on the impact of both aerosol therapy regimen characteristics and device characteristics on the process of adherence.

4.3.1 Regimen characteristics

The analysis revealed that parents and adolescents found that many of the characteristics of the aerosol therapy treatment regimen had a negative influence on the process of adhering to treatment. Particular areas highlighted were the fact that aerosol therapy was only one aspect of an already time consuming and demanding treatment regimen. Treatment regimens that are time consuming and burdensome have consistently demonstrated an association with poorer adherence in children and adolescents with a chronic health condition (Hayford & Ross, 1988; Lemanek et al., 2001; April et al., 2008) and more specifically for those living with CF (Passero, Remor, & Salomon, 1981; Modi & Quittner, 2006; Williams, Mukhopadhyay, Dowell, & Coyle, 2007; Bucks et al., 2009).

Related to the above was the novel finding that adolescents and parents felt that the interdependent nature of certain treatments – particularly between aerosol therapy and physiotherapy – added an additional layer of complexity to the process of adhering to

nebulised treatments, something not previously explored in the literature. This could be because *interdependence* is something specifically related to the process of adhering to the CF treatment regimen. However, it could also reflect the fact that *interdependence* of certain treatments (e.g., the necessity of waiting an hour after doing physiotherapy before being able to take aerosol therapy) is more commonly described within the research literature under the general heading of *complexity*. Indeed, the complexity of a given treatment regimen and the way in which this negatively impacts on rates of adherence is a widespread finding for those children and adolescents living with a chronic health condition (Glasgow, McCaul, & Schafer, 1986; Feldman et al., 2007; April, et al., 2008) and more specifically for those living with CF (Passero et al., 1981; Dziuban et al., 2010; George et al., 2010).

The quantity and type of medication prescribed as part of the aerosol therapy treatment regimen was reported as an additional factor which negatively impacted on the process of adherence. Previous research supports this observation with lower adherence being found in three times daily vs. two times daily medication regimens (Van Dyke, Lee, Johnson, Wiznia, Mohan, & Stanley 2002) for example. Related to this issue is the finding that some of the parents felt that the hospital doctors did not appreciate and empathise with the burden associated with increasing the prescribed number of aerosolised therapy treatments. Clearly, the quality of the provider-patient interaction is important with many studies suggesting that practitioners' ability to make empathic, affective contact with patients improves satisfaction with the consultation and increases the commitment to a treatment plan and overall adherence to the regimen (Becker & Maiman 1975; Squier, 1990). However, perhaps surprisingly the participants did not place much emphasis on the impact of the health providers' behaviour on the process of adherence, a finding which echoes Hall et al's (Hall, Roter & Katz, 1988) influential meta-analysis which concluded that overall, provider behaviour is relatively weakly related to adherence. This does not mean, of course that health providers do not have the potential to influence adherence, but it does indicate that other factors are far more salient to the families in this study.

Several parents and adolescents described specific unpleasant side effects associated with taking aerosol therapy. The concept that regimens which produce more negative side effects should be associated with lower adherence makes intuitive sense. Research examining this hypothesis in patients with asthma for example, found that unpleasant tasting medications were related to lower adherence (Slack & Brooks, 1995; Celano et al., 1998). However, fewer studies have investigated this factor in CF and those more recent studies which have identified a series of barriers to adherence in adolescents with CF have failed to

identify unpleasant side effects as a major difficulty (Dziuban et al., 2010; George et al., 2010; Bregnballe et al., 2011). One potential explanation for this is that the above studies examined potential barriers to adherence for the overall regimen as opposed to specific treatments. It could be argued that studies examining barriers to specific aspects of the treatment regimen are more likely to focus on participants' responses and provide more detailed information about the barriers and facilitators associated with the particular treatment under investigation. Indeed, the current study was specifically designed to attempt to achieve that aim.

One patient identified that he actually liked the taste of a particular aerosolised antibiotic that another patient found unpalatable. This was an interesting finding. However, previous studies have found that although children and adolescents may have taste preferences for different antibiotics, there is no differentially related adherence associated with these preferences (El-Charr, Mardy, Wehlou, & Rubin, 1996; Matsui, Barron, & Rieder, 1996).

Adolescent patients said that the lack of immediate benefits gained from taking aerosol therapy can negatively impact on adherence. This is a common finding within the research literature as the immediacy of benefits gained from treatments prescribed to children and adolescents living with a chronic health condition are often delayed (Lovell, Giannini, & Brewer, 1984; Rapoff, 2010).

4.3.2 Device characteristics

Parents and adolescents identified a number of characteristics of the I-neb™ device itself which had an impact on the process of adherence to aerosol therapy. Many of these characteristics were related to technological advances that were experienced as having a positive influence. Parents for example, highlighted that they were able to compare the advances of the newer I-neb™ device to those more cumbersome and demanding devices that were previously used. Parents described the associated relief of not having to endure some of the difficulties that accompanied the older devices (e.g., larger devices with more components that had to be administered with the window open to aid ventilation). Whilst the adolescents did not specifically highlight this as a motivating factor – perhaps reflecting their inability to remember the burden associated with the older devices – parents clearly felt fortunate that the I-neb™ device was smaller, less demanding and easier to use. Although nebulisers have been used for administration of inhaled medications for the treatment of a number of chronic health conditions (e.g., pulmonary infections, asthma) since the 1920s, it

was the recognition of exactly the types of limitations of conventional nebulisers that were identified by the parents that led to the development of newer devices – such as the I-neb™ which utilises AAD technology – over the last two decades (Denyer, Nikander & Smith, 2004).

The recency of the topic concerning the ways in which technological advances may influence adherence to treatment in CF serves to limit the existing research investigating the relationship. However, there have been a number of studies examining devices that utilise the AAD system compared to conventional nebulisers. These studies indicate that more complete doses are taken with the newer devices when compared with conventional nebulisers (Marsden, Conway, Dodd, & Weller, 2002), they are easier to use, administration times are reported to be significantly reduced when compared with conventional nebulisers (Mulrennan, Moon, & Morice, 2004) and patients found it better to use than their older nebuliser device (Marsden, Conway, Dodd, Edenborough, Rigby, Taylor, & Weller, 2002). In addition, there were a number of promising studies that presented initial findings at the recent European Cystic Fibrosis Society conference (Journal of Cystic Fibrosis, 2012) which indicated the potential usefulness of newer technologies in encouraging and motivating adherence to treatment (Pollard, Watson, Conway, Etherington, Ollerton, & Peckham, 2012).

The ability of TIM to provide specific feedback on breathing technique through the use of a vibrating mouthpiece was identified by both the parents and adolescents as having a positive impact on accurate breathing technique, resulting in quicker administration times. Some parents and adolescents thought the reduction in administration times were so significant that it had led to greater motivation to adhere to the I-neb™ and a reduction in oppositional behaviour and arguments. These findings are certainly in keeping with previous research with a number of studies suggesting that TIM is acceptable to patients (Denyer, Black, Nikander, Dyche, & Prince, 2010; Denyer, Prince, Dixon, Agent, Pryor, & Hodson, 2010; Goodman, Morgan, Nikander, Hinch, & Coughlin, 2010) and a more recent study finding that TIM significantly reduces treatment administration times and may positively influence adherence to treatment (McCormack, McNamara, & Southern, 2011).

Adolescents also highlighted the ability of the I-neb™ to provide specific feedback on correct positioning of the device (e.g., the use of an alarm) and completion of treatment administration (e.g., the use of an auditory beep and a visual LED smiling face). Denyer & Dyche (2010) argue that the role of feedback signals throughout nebulisation with the I-neb™ is essential to a successful drug delivery. From a psychobiological perspective, positive and negative feedback mechanisms regulate a huge amount of human physiological processes

(Carlson, 2007; Kalat, 2007). The human body is therefore not only naturally programmed to seek out and understand feedback but traditional behavioural theories such as operant conditioning (Skinner, 1948; 1953) further suggest that when a particular action has good consequences (such as positive feedback), the action will tend to be repeated whereas an action associated with bad consequences (such as negative feedback) will tend not to be repeated. A number of clinical studies have examined the use of feedback on the compliance with the correct use of devices that utilise AAD technology (Conway, Dodd, Marsden, Paul, & Weller, 2002; Nikander, Arheden, Denyer, & Cobos, 2003; Goodman, Morgan, Nikander, Hinch & Coughlin, 2010). These studies found that over 80% of patients administered the AAD device until the feedback cascade informed them that they had come to the end of nebulisation. In the current study, adolescents highlighted the role of feedback both in terms of the way it encouraged adherence to aerosol therapy and how it improved the overall accuracy of the treatment.

Related to the above issue of device feedback was the finding that parents and adolescents had mixed opinions as to the usefulness of the patient logging system (PLS) data downloaded from the I-neb™ device. Many felt that it was extremely useful whereas some were more ambivalent. This was true both for within outpatient appointments and within the current research interview process itself. In terms of clinical settings, PLS data can be actively used by clinicians to monitor their patients and identify those who are not adhering to their aerosol therapy treatment or failing to use the I-neb™ device correctly (Denyer & Dyche, 2010). A number of studies utilising this PLS data found that rates of adherence to nebulised antibiotics were generally low (50%) in adults (Latchford et al., 2009) and slightly better in children and adolescents (between 60 and 70%) (McNamara et al., 2009). The authors concluded that whilst AAD system technology may improve medication administration, rates of adherence may still be sub-optimal. They went on to suggest that clinicians can utilise PLS data to routinely monitor adherence within an outpatient setting but that they should be mindful of the pattern of non-adherence for each individual patient and the multiple factors which may be influencing this.

The way in which this data is used and communicated to patients and their parents is clearly important. The current study found that one patient had a preference for the gentle and encouraging way that her CF clinician utilised the PLS data in her outpatient appointment compared to the pejorative way in which her diabetes clinician utilised similar data downloaded from her attached insulin pump. Some approaches within the literature favour the former approach. Motivational Interviewing (MI) (Miller, 1983; Miller & Rollnick,

1981) for example, is a directive, patient-centred counselling style that aims to help patients explore and resolve their ambivalence about behaviour change. MI has been used in medical settings for a number of years and a recent review suggests that MI is well-received by patients and is an effective framework for guiding CF outpatient consultations particularly in order to encourage and improve adherence (Duff & Latchford, 2010).

Parents and adolescents also reported that certain characteristics associated with the device were burdensome. The adolescents reported that it was common for them to prepare the medication and device ready for administration whereas parents tended to be responsible for cleaning the device afterwards. Although this preparation was not specifically highlighted as problematic for the adolescents, the process was clearly very detailed and represented an additional degree of complexity when compared to treatments such as oral medications. In contrast, the process of cleaning was identified as very burdensome, particularly by the parents. It is vitally important to adequately clean the device in order to prevent harmful bacteria growing and to ensure optimal efficacy of medication delivery. The process of cleaning is quite complex (Koninklijke Philips Electronics, 2010). Parents were clearly aware of the importance of cleaning but said that it was onerous and negatively impacted on the portability of the device which made adhering to aerosol therapy outside of the home problematic. The issues highlighted may go some way to explaining why recent research indicated that approaching 40% of patients do not clean their nebuliser at all (Boyle & Hall, 2012). Patients also highlighted additional reasons for the lack of portability of the device including experiencing the device as fragile and expensive and they discussed how this negatively impacts on the process of adherence. This was an important point as on the one hand, many of the devices technological advances appeared to encourage the process of adherence but on the other hand, the resulting complications of additional cleaning and the anxiety associated with the fear of losing or damaging a device perceived as very expensive seemed to act as barriers.

4.4 Cognitive, affective and behavioural processes

Analysis revealed the complex way in which parents' and adolescents' thoughts and beliefs about aerosol therapy interacted with a series of emotional and behavioural responses which all had their relative strengths and weaknesses in facilitating the process of adherence.

4.4.1 Beliefs about aerosol therapy

Parents and adolescents reported a number of salient thoughts and beliefs concerning aerosol therapy. This is a common finding within the research literature with many studies suggesting that patients hold complex beliefs about medications and that these might influence adherence to treatment (Conrad, 1985; Britten, 1994; Horn, 1997). Although variation exists across studies in terms of the specific type of beliefs that are associated with adherence, findings indicate that certain cognitive variables included in the Theory of Planned Behaviour (TPB: Ajzen, 1988) and the Health Belief Model (HBM: Rosenstock, 1974; Janz & Becker, 1984) appear to predict adherence in certain situations. Adherence decisions for example, may be influenced by a cost-benefit analysis whereby the perceived *benefits* of treatment are weighted against the perceived *barriers* (Nelson, Stason, Neutra, Soloman, & McArdle, 1978; Cummings, Becker, Kirscht, & Levin, 1981; Brownlee-Duffeck, Peterson, Simonds, Goldstein, Kilo, Hoette, 1987). The main benefit associated with adhering to aerosol therapy as identified by both the parents and the adolescents in the current study was that the treatment is effective. However, the above research suggests that these beneficial effects are likely to be considered alongside the numerous barriers – many of which have already been discussed in detail above – and it is this cost-benefit analysis which can influence adherence.

Although it would appear that for many of the participants, the belief in the positive benefits gained from taking aerosol therapy outweighed the associated costs, some participants were less sure as to the benefit gained, arguably making the cost-benefit analysis less clear cut. In addition, patients also reported that they were influenced by the beliefs they had about preferences for different treatments. They described wide variation in the costs and benefits associated with different treatments and it was clear that this internal hierarchy of treatment preference was often responsible for influencing the process of adherence. For some patients, it is also possible that their own preferences for a particular treatment might not match their parents' preferences leading to additional complications, potential disagreements or conflict.

Further variables of the HBM and TPB are reflected in research that found higher rates of adherence in those patients who believe that failure to take the treatment could result in adverse consequences that they are personally susceptible to (Nelson et al., 1978; Cummings et al., 1981; Kelly, Mannon & Scott, 1987). The parents and adolescents in the current study reported specific beliefs about the potential consequences of not adhering to

aerosol therapy. It is likely that these thoughts and beliefs about consequences were influencing the feelings and behaviour of many of the participants and helping to encourage the process of adherence. One patient however (Karen) said that although she was aware of the consequences at a cognitive level, it did not particularly encourage her to adhere to the aerosol therapy. It is possible that for this patient, the cost-benefit analysis described was not convincing enough to drive adherence behaviour. Although it is likely that a myriad of additional biopsychosocial influences were impacting on this patient's adherence behaviour, it is interesting that her adherence rate was the lowest of all participants, at 28%.

Many of the above concepts relate to more systematic research by Horne and colleagues (Horne, 2003; Horne, Weinman & Hankins, 1999) that suggest that four core themes – *specific necessity*, *specific concerns*, *general harm* and *general overuse* – underlie commonly held beliefs about medications. The first two factors describe people's beliefs about their prescribed medicines and were found to be more strongly related to adherence reports than the second two factors which describe people's beliefs about medicines in general. *Specific necessity* describes beliefs that people have around the perceived need of the prescribed medication for maintaining health. Adolescents in the current study reported beliefs they held about the necessity of adhering to the aerosol therapy medication. Many of these beliefs related to the way in which they perceived their symptoms, often linking a greater necessity to adhere to aerosol therapy with feeling physically unwell. Previous studies have shown that those with stronger beliefs in the necessity of their prescribed medication reported greater adherence to the regimen (Horne, 2003; Horne et al., 1999). This research also showed that, although illness perceptions (such as the way the adolescents perceived the severity of their symptoms) can explain variance in adherence, they may play a more important role in determining beliefs about treatment. In a study of factors influencing adherence to preventative treatments for patients with asthma for example, stronger beliefs about the necessity of taking these medications were found in those who perceived their condition as more chronic with greater negative consequences (Horne & Weinman, 2002).

There are other theories with a focus on cognitive approaches to non-adherence. Kendall (1993) argues that thoughts or self-generated rules can influence adherence behaviour in two general ways. Firstly, patients and parents can fail to generate thoughts or rules about the health condition and associated treatment regimen when it would be helpful to do so (e.g., "I need to take my treatment consistently in order to give the medication a chance to work."). Secondly, patients and parents may actually generate counterproductive thoughts or rules (e.g., "I'll take my treatment so long as I'm not too busy"). The current study

provided examples of both the failure to generate cognitions (e.g., Karen: “*I’ve been told the consequences and stuff of not taking it and I still didn’t listen*”) and the generation of unhelpful cognitions (e.g., Lindsey: “*You tend to think, ‘well you’ll be alright for a few days’ or whatever, ‘it’s only one treatment’*”) that were related to the process of adhering to aerosol therapy.

Leventhal’s self-regulatory model (Leventhal, 1993; Leventhal et al., 1984) is a further example of a dynamic cognitive approach which has been utilised in the explanation of non-adherence. Marteau & Weinman (2004, pg.287) suggest that the model “acknowledges the importance of symptom perception in influencing illness representations which, in turn, direct coping responses, including adherence behaviour.” Research has linked illness perceptions with a variety of behaviours that influence the process of adherence. In a study examining the role of illness perceptions and treatment beliefs in adolescents with CF for example, Bucks and colleagues (Bucks et al., 2009) reported that doubts about the necessity of antibiotics and a belief that CF is not able to be controlled by treatment were both related to non-adherence.

The above theories and models have been formulated and tested mostly within adult populations so, by necessity, many of the studies referred to above have been extrapolated from the adult literature in order to apply these theories to children and adolescents. The main exception to this is the HBM, which *has* been adapted for use within paediatric populations. The Children’s Health Belief Model (CHBM: Bush & Iannotti, 1990) not only includes very similar variables as the traditional HBM (e.g., perceived benefits and perceived barriers) but also places additional emphasis on the role of parental influences on children’s health beliefs and actions (e.g., parental perceived benefit of the child taking prescribed medication). What was clear within the current study was that parents (as well as adolescents) held complex thoughts and beliefs about aerosol therapy and that these not only had an influence on their own emotions and behaviour, but those of their children.

Although not adapted specifically for paediatric populations, the TPB also emphasises that the perceived views of significant others, such as family and friends (*normative beliefs*) may also influence adherence (Cochrane & Gitlin, 1988; Conner & Sparks, 2005). This would suggest that the adolescents in the current study were more likely to adhere to the aerosol therapy where parental beliefs in the benefit of taking the treatment were strong. However, it was clear from the parental reports that their beliefs were not necessarily static as some parents expressed having doubts about the effectiveness of aerosol therapy from time-to-time, particularly when their child became unwell.

Even where beliefs in the treatment were consistently strong, many of the parents also held beliefs about the unlikelihood that adherence to aerosol therapy would always be one hundred percent. They felt that there were times when the many daily barriers to adherence (e.g., lack of time, having to clean the device or their child feeling tired) outweighed the benefits. Rapoff (2010) argues that this reality of non-adherence reflects “part of a mosaic of patient and family struggles” (pg.184) which makes the prospect of consistently perfect adherence rates very unlikely. Whilst advances in the efficacy of treatment for CF may have encouraged patients and parents to believe in the value of taking medications such as aerosol therapy, there are broader psychosocial contexts that are likely to continue to impact on non-adherence.

4.4.2 Emotional drivers

Parents reported a number of emotions associated with the process of their child adhering to aerosol therapy. These were often described in relation to the way that they interacted with thoughts and beliefs and subsequently initiated certain behavioural patterns. One area that caused parents particular concern was the uncomfortable feelings generated at the thought of relinquishing the control they had over their child’s adherence to CF treatments. Most of the parents, for example, described having taken full responsibility for their child’s adherence to aerosol therapy for many years (e.g., ordering the medication, preparing the treatment, handing their child the device, supervising them taking it and then cleaning the device afterwards) and said that the idea of this responsibility being passed over to their child was experienced as difficult. The struggle to find a balance between greater autonomy for the adolescent and assurance that daily CF treatments will be adhered to has been previously reported in the literature (Delambo, Ievers-Landis, & Drotar, 2004; Fiese & Everhart, 2006; Smith & Wood, 2007). What was clear within the current study was that attempts to relinquish control over adherence to aerosol therapy caused significant anxiety for some parents and led to increased monitoring and supervision through the use of frequent daily reminders. Linked to this issue was the finding that some parents often had feelings of guilt when their child had not taken their aerosol therapy, particularly if they had failed to remind their child to take it. It would appear that the use of certain behavioural strategies (such as frequent reminders), were not only employed to encourage their child to adhere to aerosol therapy so that they secured the associated health benefits, but also to relieve their own uncomfortable feelings of anxiety and guilt.

Difficulties in relinquishing control were not the only sources of anxiety and worry reported by the parents. Being anxious about the disease itself, the child's health status and general treatment concerns (e.g., worrying whether enough medication had been ordered) were all potential causes of negative affect. Increased levels of anxiety are not uncommon in those parents caring for a child with CF (Barker & Quittner, 2010) and greater levels of parental stress and anxiety have been associated with lower levels of adherence to treatment in other chronic health conditions (Hazzard et al., 1990; Brownbridge & Fielding, 1994; Gerson et al., 2004). There were parents in the current study who also described feelings of frustration towards some of their child's behaviour in relation to the process of adhering to aerosol therapy. The child forgetting to take the aerosol therapy, getting distracted, procrastinating or failing to breathe correctly when using the I-neb™ device were all examples of sources of frustration which had the potential to impact negatively on their day-to-day interactions with their child. Although understandable, these types of negative parent-child interactions have been associated with lower adherence to treatment regimens for renal disease (Gerson et al., 2004), diabetes (Miller & Drotar, 2007) and CF (Quittner, Drotar, & Ievers-Landis, 2004).

It is clear from the findings of the current study that parents caring for a child with CF are under significant pressure. It is perhaps unsurprising therefore, that higher rates of depression have been found in those parents caring for a child with CF (Snell, Barker, & Marciel, 2008; Yilmaz, Sogut, & Gulle, 2008). In addition, emerging evidence suggests that parents may experience "burnout" in their role as caregiver during adolescence which highlights the importance of monitoring both children and parents' psychological and emotional distress and providing additional support to parent caregivers where appropriate (Snell et al., 2008; Barker & Quittner, 2010; Smith, Modi, Quittner, & Wood, 2010). Intervening in this way is particularly important when taking into consideration that a higher level of parental stress and depression is associated with lower levels of adherence to treatment (Quittner, Barker, & Geller, 2007; Ward, Massie, & Glazner, 2009).

The adolescents in the current study described the consequences resulting from some of the more negative aspects of the parent-child relationship. Patient experiences of their parent or other members of their family getting frustrated as a result of them forgetting to take the aerosol therapy contributed to feelings of not being understood. Reports indicated that this contributed to their *own* feelings of frustration and a sense of being different to others. Lower levels of adherence in CF have been linked to increased conflict with parents, time spent outside of the home and away from parental monitoring and a heightened desire to

be similar to friends and peers who are healthy (DeLambo et al., 2004; Badlan, 2006; Smith & Wood, 2007; Modi et al., 2008). A reluctance to disclose their illness to peers and an associated unwillingness to take their treatment in front of others have all been previously reported in the literature (D'Auria, Christian, & Richerdson, 1997; George et al., 2010; Bregnballe et al., 2011). However, the finding that adolescents in the current study felt embarrassed taking their aerosol therapy in front of their parents as well as their peers in certain situations (e.g., when parents are watching the TV) and the finding that adolescents experienced adhering to some types of treatment harder than others when in public (e.g., nebulisers were harder than tablets because tablets were less time consuming and more portable) were novel. Although few studies have examined how close friends and peers can influence adherence, it was clear that for some of the adolescents certain types of physiotherapy (e.g., trampolining) were more amenable to involving friends, therefore serving to encourage adherence, than treatments such as aerosol therapy.

Managing the burden of living with a chronic health condition can take its toll on children and adolescents. Children experiencing emotional difficulties are less likely to adhere to treatments prescribed for diabetes, cancer and renal disease (Brownbridge & Fielding, 1994; Kennard, Stewart, Olvera, Bawdon, Ailin, & Lewis, 2004; Greening, et al., 2007). The simple fact that adolescents may feel tired and decide to not take their treatments was reported in a study which examined the barriers to adherence in CF (George et al., 2010). The adolescents in the current study echoed the above findings in describing how feeling upset or tired negatively impacted on the process of adherence to aerosol therapy. Whilst fearing the consequences of non-adherence was not a pleasant experience, the adolescents reported that this often helped to *encourage* adherence and is consistent with recent findings that investigated facilitators of adherence behaviour in CF (George et al., 2010).

Although none of the adolescents in the current study reported feelings of significant depression, the impact of negative affect on adherence is a growing area of research interest. The International Depression and Anxiety Epidemiological Study (TIDES) was established to conduct the first international systematic research investigating anxious and depressive symptoms in young patients with CF and their parent caregivers (Quittner, 2012). The TIDES data indicates that 25% of adolescents with CF reported depressive symptoms above a clinical cut-off, with 20% reporting anxious symptomology. These are important findings when considered alongside recent evidence which indicates that more depressive symptoms in children and adolescents with CF are related to poorer rates of adherence (Smith et al., 2010). Although the routine monitoring of depression and anxiety is not commonplace

within CF outpatient clinics (Barker & Quittner, 2010), the routine monitoring of aerosol therapy adherence data has become much more frequent (McNamara et al., 2009). Through careful and sensitive exploration of objectively poor levels of adherence to aerosol therapy, clinicians have the opportunity to help to ascertain if negative affect – experienced by either adolescents or parents – is impacting on the process of adherence. If so, offering appropriate levels of support or more specialist psychological interventions may help to address these issues and improve adherence behaviour.

4.4.3 Strategies and behaviours

Several strategies and behaviours related to the process of adherence were reported by the parents and adolescents. The way in which a well defined and regular routine – particularly during the school-week – helped to facilitate adherence to aerosol therapy was highlighted as important by virtually all of the participants. Even when people hold positive beliefs about treatment and have intentions to adhere, Abraham and colleagues (Abraham, Sheeran & Johnston, 1998) acknowledge that intentions do not automatically translate into action and suggest that it is prior planning and subsequent rehearsal that can help to enhance individual control of action and facilitate the routinisation of health-related behaviour. The participants in the current study were clear that a well planned and rehearsed routine was vitally important, with some saying that a lack of routine was a barrier to adhering to aerosol therapy. This finding is very common in the literature, with a number of studies consistently highlighting that a lack of routine is a barrier to adherence in CF (Dziuban et al., 2010; George et al., 2010). Routinisation of adherence was further reflected in the parental reports of normalising and accepting that adhering to aerosol therapy was part of life and needed to be effectively planned and managed. Adolescent reports of “getting it done and out of the way” were an additional behavioural strategy that related to routine and acceptance.

One strategy for improving the planning and rehearsal necessary to facilitate the routinisation of health-related behaviour is to devise and agree upon an implementation plan (Gollwitzer, 1999). *Implementation intentions* are self-regulatory strategies which utilise an *if-then-plan* (i.e., “If situation X arises, I will perform response Y”) and are used to increase the likelihood of attaining goals. A meta-analysis showed that implementation intentions had a positive effect ($d = .65$) on goal attainment and were effective in promoting the initiation of goal striving and the shielding of goal pursuit from unwanted influences. Although not specifically investigated for use in the encouragement of adherence to treatment in CF, implementation intentions are clearly helpful and could be utilised in clinical settings

for this purpose. Research which examines the effectiveness of implementation intentions used to encourage adherence in CF may be warranted.

Parents said that they frequently used reminders as a way of monitoring and supervising their child's adherence to aerosol therapy. There are studies which found that lack of parental monitoring of treatment behaviour and activity can contribute significantly to non-adherence, particularly to chronic disease regimens (Wiebe, Berg, Korbel, Palmer, Beveridge, & Upchurch, 2005; Rapoff, 2010). One study examining the management strategies of children with diabetes found that parental monitoring and supervision had all but ended by the time the children had reached 15 years of age (Ingersoll, Orr, Herrold, & Golden, 1986). In CF, Modi and colleagues (Modi et al., 2008) found better adherence in adolescents who spent greater amounts of their treatment time supervised by their parents, particularly mothers. However, there was evidence in the current study that the patients may not appreciate having to spend increased periods of time being supervised by their parents, particularly with the older adolescents who enjoyed an active social life outside of the family home. These conflicted views and resulting behaviour may have the potential to negatively impact on the parent-adolescent relationship.

Whilst reminding their child to take their CF treatments appears to be a popular strategy for parents (Cowlard, Yorke & Carr, 2010) and can sometimes help to facilitate adherence (George et al., 2010) it is not necessarily the most effective way of monitoring behaviour and patients in the current study had mixed views about how helpful they found them (i.e., on the one hand they helped to prompt adherence behaviour but on the other hand, they were experienced as frustrating). General reminders about treatment were described by some of the parents in the current study as a way of attempting to alleviate the anxiety associated with fears about their child's non-adherence. A study by Ellis and colleagues (Ellis, Yopp, Templin, Naar-King, Frey, & Cunningham, 2007) found that comprehensive diabetes-specific, rather than more general parental supervision and monitoring was associated with higher adherence. Rapoff (2010) suggests that the use of monitoring charts (e.g., where parents and children can "tick off" when a particular treatment has been completed) and checking medication supplies (e.g., vials of medication) and devices (e.g., nebulisers or blood glucose meters) for indirect evidence of adherence are all potentially useful ways of ensuring adequate supervision alongside more general strategies such as the use of prompts. However, he further argues that the monitoring and supervision of treatment regimens needs to be done in a way that is sensitive to the needs and developmental capacities of the child. The use of individualised and gentle reminders, alongside a series of

other monitoring and encouragement strategies, were said to be helpful for two of the adolescent patients in the current study.

One can understand why reminders feature so heavily in the daily interactions between parent and child when taking into account the literature which suggests that children simply forgetting to take their medication is one of the most common barriers to adherence in CF and other chronic health conditions (Shemesh et al., 2004; Modi & Quittner, 2006; Dziuban et al., 2010; Bregnballe et al., 2011). Monitoring strategies such as those described above and pairing regimen tasks with regularly occurring daily events to aid routinisation (e.g., taking aerosol therapy before the evening meal), may help to prompt the type of non-intentional forgetting that is frequently described within the literature (Park & Kidder, 1996) and also featured in the adolescent and parent reports.

However, there were participants who described more purposeful forgetting which was used as a way to prevent them from being reminding of the fact that they had CF. This type of purposeful forgetting can be a significant barrier to adherence in CF (George et al., 2010). In a study examining coping and CF, Abbott (2003) suggests that omitting treatments can be an attractive option for those who are keen to escape the reality of the condition. The patients' accounts describing "putting off" taking their aerosol therapy can perhaps be understood in a very similar way. This type of behaviour is not particularly well understood and can often be dismissed as the child or adolescent simply "being difficult". Whilst traditional theories have attempted to understand adherence behaviour in terms of more conscious, cognitive processes (e.g., the role of beliefs, perceptions and attitudes), psychodynamic approaches posit that less conscious processes may be influencing adherence. Alfonso (2011) for example, suggests that traditional psychoanalytic defences such as denial (e.g., refusing to acknowledge the painful reality of being diagnosed with a chronic health condition), acting out (e.g., acting unacceptably to give expression to the emotional distress associated with living with a chronic health condition) and passive aggression (e.g., unassertively expressing aggressive feelings towards those involved in their care by overtly complying with their treatment advice while covertly resisting this through non-adherence) may help to provide a different level of understanding regarding complex adherence behaviours. Systematic research into this intriguing area is limited, although there are some good examples of smaller studies which have examined these concepts in the context of non-adherence to chronic health treatment regimens in older adolescents and adults (Ciechanowski, Katon, Russo, & Walker, 2001; Ricart, Cohen, & Alfonso, 2002). While their child forgetting to take their aerosol therapy was a source of frustration for the parents in

the current study, Cowlard et al (2010) suggest that it is important for parents and clinicians to understand “forgetting behaviour” – particularly purposeful forgetting – in the context of children and adolescents attempting to achieve a normal lifestyle and try to allow for individuality and flexibility as opposed to seeing it simply as a form of rebellion.

Some of the earliest research into the determinants of non-adherence hypothesised that inadequate understanding of the illness and treatment would lead to incomplete adherence (D’Angelo & Lask, 2001). Parents in the current study felt that it was vitally important that their children understood the purpose of adhering to their aerosol therapy and described taking an active role in helping to facilitate this. Although it is not always the case, research has tended to demonstrate that children and adolescents who are less knowledgeable about their illness and treatment are less likely to adhere to regimens for diabetes, CF and cancer (Tebbi et al., 1986; Gudas, et al., 1991; Holmes, et al., 2006). Whilst this may be true, it is important to acknowledge that increased knowledge about treatments does not always translate into improved rates of adherence (McQuaid, et al., 2003). Although it was clear from the interviews that patients and parents were very knowledgeable about the CF treatments, analysis revealed that there were numerous additional influences on the process of adhering to aerosol therapy.

Parents in the current study said that they offered a number of additional supportive strategies to attempt to encourage adherence. Simplifying the process of adhering to aerosol therapy was a common strategy identified by the parents as was rewarding and incentivising adherence. The use of rewards and incentives to encourage adherence has been frequently examined within the literature. Token reinforcement programmes (whereby parents provide or confiscate points or chips for adherence which can then be used to “purchase” special privileges) have demonstrated positive effects on improving adherence to chronic illness regimens (Pieper, Rapoff, Purviance, & Lindsley, 1989; da Costa, Rapoff, Lemanek, & Goldstein, 1997). More simplistic strategies such as parents contracting with their children about how they will be rewarded for consistent adherence and also what consequences will occur as a result of non-adherence can have a similar effect, particularly if communicated appropriately and applied on a consistent basis (Rapoff, 2010). Reports from both parents and adolescents in the current study suggested that these types of contracts – which provide rewards and incentives for taking aerosol therapy – are commonplace and often helped to facilitate adherence.

In addition, adolescents were also responsible for more self-generated incentives for adherence; describing for example, how getting their aerosol therapy done quickly resulted in

increased free time and allowed them to engage in more enjoyable activities. This is consistent with the findings reported by George and colleagues (George et al., 2010) who suggested that self-generated reward systems can help to facilitate adherence in adolescents with CF. A further example could be found in patient accounts of trying to breathe slowly and appropriately in order to ensure accurate administration of the aerosol therapy, which, paradoxically helped to speed up treatment times.

Parent and adolescent beliefs about the unlikelihood that adherence to aerosol therapy would always be one hundred percent were discussed earlier. There was certainly evidence in both the parent and adolescent accounts that some of these beliefs translated into actual behaviour. Having either a planned or more spontaneous break from adhering to aerosol therapy was at times described as very likely. This created mixed emotions in the parents whereby feelings of guilt about their child not adhering to the treatment coexisted with feelings of relief, particularly about not having to clean the device. The adolescents seemed less concerned and mostly described having a break from adhering to aerosol therapy as pleasurable, because it allowed them greater freedom. Dziuban and colleagues (Dziuban et al., 2010) found that adolescents' believed it was acceptable to miss a treatment every few days or when they were busier.

Although as clinicians, it can be difficult to accept that treatments which keep patients well are sometimes not taken, it would seem that there are times when non-adherence to treatments such as aerosol therapy is going to be very likely. Whilst it is imperative that treatments are taken in order for their benefits to be realised, Rapoff (2010) suggests that the importance of adherence is sometimes inflated. He argues that it is not only important to recognise that adherence problems may be symptomatic of broader psychosocial difficulties or family dysfunction that require appropriate psychological support, but that the outcome of medical treatment does not depend entirely on adherence. He goes on to suggest that although sub-therapeutic drug assays may reflect a patients' poor adherence, due attention should also be given to issues that are not dependent on patient behaviour, such as inadequate dosing, interactions with other drugs and pharmacokinetic variations in drug metabolism (i.e., the way peoples bodies metabolise specific treatments differently).

4.5 Parent and adolescent relationship

The day-to-day interactions between parent and adolescent and the way they experienced each other's behaviour – particularly in relation to the process of adhering to aerosol therapy – was interesting. Recall how some of the adolescents' behaviour in relation

to the aerosol therapy (e.g., forgetting to take it, getting distracting, procrastinating and inappropriate breathing technique) was a source of frustration for the parents. Similarly, some aspects of the parents' behaviour (e.g., the use of constant reminders, a lack of understanding regarding the treatment burden) had the potential to frustrate the adolescents. These irritations, particularly when considered alongside the complex and interacting beliefs and emotions experienced by the parents and adolescents, had the potential to lead to conflict. Increased conflict and negative parent-child interactions have all been associated with poor adherence to treatment in CF (DeLambo et al., 2004; Quittner et al., 2004; Badlan, 2006; Smith & Wood, 2007; Modi et al., 2008). What was clear from the analysis of parent and adolescent accounts within the current study was that although there were sources of daily frustration related to the process of adhering to aerosol therapy, the strength of the parent-adolescent relationship had the potential to act as a "buffer" that mediated between frustrating behaviour and conflicts. This is certainly in chorus with previous research which suggests that the single most consistent predictor of positive adjustment and good mental health throughout adolescence is the quality of the parent-child relationship (Steinberg & Silk, 2002)

Managing the potential for conflict in the difficult adolescence period of development can be difficult for healthy children and their parents, but presents unique challenges for those living with CF. Adolescence is an intense period of physical and pubertal development, characterised by rapid social and emotional transformation and a thrust toward autonomy (Berk, 2006). The struggle to find a balance between greater autonomy for the adolescent and assurance that daily CF treatments are being adhered to was a feature both in these parent and adolescent accounts and within the previous research literature. There was considerable variation in the parent and adolescent accounts detailing the amount of responsibility taken for the process of adhering to aerosol therapy. The younger adolescents had less responsibility, which is consistent with findings in the literature that suggest parental monitoring and supervision is greater during this phase (Fiese & Everhart, 2006). There was certainly an acknowledgement from some of the adolescents that they needed to take on more responsibility and although this was beginning to happen, particularly in terms of aerosol therapy treatment preparation, this was not always the case. Parents were still largely responsible for ordering the medication, reminding their children to take the aerosol therapy and cleaning the device afterwards.

Barker and Quittner (2010) argue that a reluctance to transfer responsibility may result in adolescents failing to learn the necessary skills to independently manage their

disease. They go on to suggest however, that a premature transfer of responsibility and a reduction in adequate monitoring and supervision may result in the adolescent becoming overwhelmed and non-adherent. It is clearly a difficult balance. It has been suggested that the benefit of parental supervision may be dependent on the quality of the parent-adolescent relationship (Smith & Wood, 2007). It was clear from one adolescent account (Elizabeth) that her ability to take on an impressive degree of responsibility for the management of her CF treatments was appropriate for her level of development and encouraged by a series of support mechanisms provided by her parents. Barker and Quittner (2010) suggest that this type of *supported autonomy* is in many ways the optimal strategy: “providing adolescents with increasing responsibility for disease management, predicted on their ability to organise their time and perform these tasks.” This model of cooperative parenting is described by Maccoby (1984) as *coregulation*, whereby parents gradually shift responsibilities and exercise oversight while at the same time permitting children to be in charge of moment-by-moment decision making. In contrast to this model was one parent account (Mary) who suggested that she had absolved virtually all responsibility for the process of adhering to aerosol therapy to her daughter: “*Like the I-neb, that’s up to her to do it ... The responsibility is on her now you know.*” Recall that, interestingly, her daughter’s adherence rate was the lowest of all participants (28%).

Further evidence of the strength of the parent-adolescent relationship was found in many of the adolescent and parent accounts detailing the way that appropriate empathy and support was provided by the parents to their child. The adolescents described how this practical and emotional support helped to facilitate adherence. In general, support of this kind has been found to increase adherence across all illnesses and ages (DiMatteo, 2004). Again, the balance appears difficult with some studies suggesting that parents were both sources of positive (helpful and appropriate reminders) and negative (drawing attention to the illness, nagging etc) behaviours (Graetz, Shute & Sawyer, 2000; Barker, Cohen, & Driscoll, 2008). Recall how the adolescents experienced reminders as helpful at times, whilst annoying at others, with one patient suggesting that it was the way in which reminders were used and communicated that enabled her to experience them as supportive. Effective communication was obviously important to the participants in the current study, with all of the adolescents reporting that having clear and consistent instructions from their parents when it came to the process of adhering to aerosol therapy was experienced as helpful. Many of them also said that it was frustrating when this was not the case. Indeed, studies examining poor communication between parents and children with chronic health conditions found that

it contributed to lower levels of adherence (Gerson et al., 2004; Lewandowski & Drotar, 2007; Miller & Drotar, 2007).

Related to the necessity of effective communication were reports by the participants detailing how cooperating and negotiating with each other helped to facilitate the process of adhering to aerosol therapy. These skills were also found in some of the parent and adolescent reports of having to overcome the specific problems associated with the process of adhering to aerosol therapy (e.g., how the aerosol therapy could be taken when the parent and child were attending a party). Better communication and problem solving between parents and children have been associated with higher rates of adherence to a number of chronic health condition treatment regimens (Bobrow et al., 1985; McQuaid et al., 2005). Linked to this were the parent accounts in the current study that suggested the importance of recognising and accommodating specific child preferences – particularly when it came to the process of adhering to aerosol therapy (e.g., child not wanting to take their aerosol therapy when coming home from school) – as a way of anticipating and managing potential problems and conflict. These supportive elements of the parent-adolescent relationship were strengthened by incentives to encourage adherence to aerosol therapy, most of which were discussed in detail earlier.

4.6 The context of adherence

Studies of “context” in relation to health and illness are becoming increasingly widespread (Frohlich, Corin, & Potvin, 2001). Although fewer studies have examined the impact of social and environmental influences on adherence to treatment in chronic health conditions, it was clear from the analysis in the current study that the context in which adherence to aerosol therapy was taking place was experienced as extremely important for the parents and adolescents.

4.6.1 Everyday life

Parent accounts of day-to-day life were characterised by attempts to fit multiple tasks (some CF treatment related, some not), for multiple people (child with CF, other children, partners, friends and colleagues) into a limited amount of time. In this way, parents seemed to be describing a type of “juggling act” that included the necessity to adhere to aerosol therapy as only one of the numerous commitments that needed to be managed. Many of the parents held beliefs about the unlikelihood that adherence to aerosol therapy would always be one hundred percent, due in no small part to exactly the type of hectic everyday lifestyle

described above. Parents and adolescents described a healthy involvement in numerous social and recreational activities, alongside more commonplace commitments such as attending school or work. Perhaps unsurprisingly, increased family social and recreational activities outside of the home have been associated with lower rates of adherence to CF treatment regimens (Patterson, 1985; Geiss, Hobbs, Hammersley-Maercklein, Kramer, & Henley, 1992). One explanation for this finding is that because families with CF children have a finite amount of time, resources and energy to devote to treatment regimens, increased recreational activities, however positive, consumes a large portion of that time. There are some who have argued that clinicians need to be mindful not to overburden families by prescribing unnecessarily complex regimens which prevent the ability to enjoy recreational activities and the maintenance of some semblance of a “normal” family life (Patterson, 1985; Rapoff, 2010).

Related to the above was the finding in the current study that parties or special occasions negatively impacted on adherence to aerosol therapy. Parents and adolescents described adherence taking on less of a priority at these times and provided the opportunity for either planned or more spontaneous non-adherence. However, it would seem that the interacting cognitive (e.g., belief in the efficacy and necessity of aerosol therapy) and emotional (e.g., feelings of anxiety and guilt) processes of some of the parents led to the instigation of specific behavioural responses (e.g., reminding their child about the need to take aerosol therapy) which were mediated by the strength of the parent and child relationship (e.g., cooperating and problem solving in order to agree upon a solution) and enabled the facilitation of adherence to aerosol therapy despite the fact that they were attending a party or special occasion. For others who were faced with the prospect of a party or celebration, it appeared that a variation or breakdown of one or more of the interacting processes described above led to non-adherence.

A similar explanation could be used to describe how some adolescents managed to adhere to aerosol therapy despite being faced with numerous lifestyle barriers, whereas others found adherence more difficult to achieve. There were some adolescents for example, who said that fun activities negatively impacted on the process of adhering to aerosol therapy whereas others were able to use the prospect of fun activities to motivate adherence. Having said that, it appeared that as well as observed differences *between* participants (i.e., in the way that these barriers were successfully negotiated), there were also differences within the *same* participants at any given time, suggesting that these interacting processes were not static and had the potential to change on a regular basis. In this way, adherence to aerosol

therapy can be understood along similar terms as numerous other health behaviours (e.g., attempting to eat a healthy diet, take more exercise etc) which also have a social and environmental component as well as a biological and psychological one.

The discovery by McNamara and colleagues (McNamara et al., 2009) that evening adherence to nebulised antibiotics was significantly better than morning adherence was novel. There were a number of interesting findings in the current study which helped to further elucidate this claim. Firstly, the majority of parents felt that it was much easier for their child to take their aerosol therapy in the evening because the morning schedule was just too busy. There was a caveat to this though, with two parents highlighting that patterns of morning and evening adherence were often dependent on the number of nebulised antibiotics prescribed. Secondly, while half of the adolescent patients agreed with their parents about evening adherence, the other half felt that morning adherence was easier. Interestingly, their perception of morning adherence being easier was not supported by the PLS data downloaded from the I-neb™ device which suggested that aerosol therapy was more likely to be taken in the evening. One patient realised this and said that he just “thought” it would be easier to take in the morning, whereas the other two patients were more surprised. There are a number of potential explanations for this. It is possible, for example, that the younger adolescent’s preference for adhering to his aerosol therapy in the morning was superseded by his parent’s predilection for evening adherence. While this may also be true for the other two adolescents, they were more surprised at the finding which may suggest that although their perception was that they thought that morning adherence was easier, the use of objective data provided them with a more accurate representation of day-to-day life.

The finding that participants’ perception of their own adherence behaviour is different to more objective measurements is consistent with the existing research literature. Gordis and colleagues (Gordis, Markowitz, & Lilienfeld, 1969) carried out one of the earliest studies that directly compared patient and parent reports of adherence with more objective measurements (e.g., urine assays) and concluded that self-reports of adherence were “grossly inaccurate”. A number of more recent studies have consistently corroborated this finding, reporting that adherence rates are lower as measured by electronic monitors vs. parent and patient report (Bender, Wamboldt, O’Conner, Rand, Szeffler, & Milgrom, 2000; Butz, Donithan, Bollinger, Rand, & Thompson, 2005; Modi et al., 2006). The findings from the current study suggest that although parents and adolescents perceive that it is easier to take their aerosol therapy at particular times, this does not necessarily translate into actual behaviour. This again highlights the usefulness of technology such as the PLS data in its

ability to be used routinely in order to provide a more accurate and objective view of adherence behaviour.

Related to the above were the reports from parents and adolescents regarding their perceptions of school day vs. weekend adherence to aerosol therapy. Essentially, preferences were evenly matched with approximately half of the parents and adolescents saying that school day adherence to aerosol therapy was easier and the other half saying that it was easier at the weekend. Although school days were busier, the planned and regular routines that were in place (particularly the after school and evening routines) were said to help facilitate adherence. In contrast, approximately half of participants felt that weekend adherence was easier because they were more relaxed and more time was available. Again, of interest was the finding that participants' perceptions of ease did not necessarily translate into actual adherence behaviour. There were instances for example, of participants' saying that they felt it was easier to adhere to aerosol therapy at the weekend when the PLS data suggested that adherence was lower at these times.

4.6.2 Environmental influences

We have considered a number of lifestyle factors identified by the participants as influencing the process of adherence. Although "lifestyle factors" have been increasingly studied in relation to their influence on a number of public health priorities (e.g., obesity, smoking etc), Frohlich and colleagues (Frohlich et al., 2001) suggest that these studies have a tendency to discuss the term in reference to individual behavioural patterns that affect disease status (e.g., Bandura 1984). They go on to argue that this has created a type of *behavioural determinism* whereby so-called *health behaviours* (e.g., eating healthy food or adhering to medication) are studied "independently of the social context, in isolation from other individuals, and as practices devoid of social meaning." What was clear from the findings in the current study was that the participants' lifestyle was intertwined with the social and environmental context within which they operated and that these closely connected phenomena had very real influence on the process of adhering to aerosol therapy.

The way in which aspects of the home environment acted as a barrier to adherence, for example, was of particular interest. The adolescents described the way that interacting with technological devices within the home (e.g., Computer, social networking sites, the TV, mobile phones etc) often led to them forgetting to take their aerosol therapy or distracted them part way through the administration of the medication. Although relatively little is known about the way characteristics within the home environment contribute to or impede

adherence, a recent study found that social demands (such as sitting and watching TV or playing on the computer) often acted as a barrier to adherence for adolescents with CF (George et al., 2010). One of the variables within the HBM (Janz & Becker, 1984) describes the way that *cues to action* within a person's environment can help to enhance or compromise health behaviour. Parents within the current study said that the visibility of the I-neb™ device and associated medication and equipment helped to normalise aerosol therapy treatment but did not necessarily facilitate adherence. Although studies are beginning to investigate in more detail the impact of the home environment on adherence to treatment (e.g., Vaughan, Wagner, Miyashiro, Ryan, & Scott, 2011) much remains unknown about this interesting area.

Participants said that there were a number of difficulties associated with taking the I-neb™ device outside of the home environment. It is perhaps unsurprising therefore, that both parents and adolescents highlighted aspects of the external environment that they felt negatively impacted on the process of adhering to aerosol therapy. Going away on holidays or school trips was particularly difficult, largely because of the problems identified earlier around having to clean the device thoroughly and store all of the associated medication in a fridge. Interestingly, being admitted to hospital caused the same difficulty which is surprising considering the recent emphasis on providing appropriate washing facilities as part of the NHS infection control strategy (NHS Professionals, 2010). Staying over in a different house was a similar situation, although there were additional complications for those adolescents who were reluctant to take their aerosol therapy in front of their friends (discussed earlier) and those whose parents were separated. Having to take the I-neb™ device and associated medication and equipment over to another parent's house when the parents were separated was identified as burdensome. In addition, one parent highlighted that she did not allow her son to take his aerosol therapy with him to his father's house in case he forgot to bring it back, resulting in daily non-adherence once a week. A similar problem was fortuitously solved for one of the other participants by having two I-neb™ devices (i.e., she was given a new device when her original one malfunctioned and then was then given the repaired one back to use as a spare). It was decided that she would keep one device at her father's house and one at her mother's because it made the process of adherence much easier. Although this is a very specific incident related to divorce, more general research has found that parental separation and divorce is associated with lower adherence to treatments in a number of chronic health conditions (Brownbridge & Fielding, 1994; Shemesh et al., 2004). Clinicians may wish to sensitively explore any specific adherence concerns related to the

issue of parental separation (such as those discussed above) so that appropriate support can be given or potential solutions to problems can be identified.

Finally, although greater father involvement (e.g., amount and helpfulness) has been related to better adherence to treatment in CF (Wysocki & Gavin, 2006), it has been consistently demonstrated that mothers take on the majority of responsibility for their child's adherence to treatment (Rapoff, 2010). This was both anticipated and realised within the current study.

4.7 Overall summary

The results of the current study complement and extend previous research on adherence in CF. In particular, it has highlighted the complexity involved, and the interactions which have a massive influence on adherence – interactions between the different parts of the treatment regimen, interactions between the nebuliser and child and nebuliser and parent, interactions between children and their peers, and finally between child and parent. It has also placed adherence behaviour in context – the internal context of the beliefs and emotions of the child and parent, and the exterior context of the busy and complicated lives led by them. It would appear that these interacting biopsychosocial processes make the prospect of consistent, one hundred percent adherence to aerosol therapy very unlikely.

4.8 Methodological considerations

The following section will summarise the methodological considerations of the current study. Firstly, the limitations of the study will be highlighted, before moving on to consider the study's strengths.

4.8.1 Methodological limitations

4.8.1.1 Researcher bias

Some level of interpretation and imposed structure to aid the manageability of large quantities of data is considered inevitable in qualitative research (Smith, 2008). However, this creates an added emphasis on the necessity of the researcher to bring into conscious awareness their own unique biases and the ways in which these have the potential to influence the process of data analysis (Bryant & Charmaz, 2007). Section 2.9.3 outlined the chief investigators previous experiences, clinical and theoretical influences and emotional reactions to the current participants in an attempt to highlight sources of potential bias.

Of these, the fact that the chief investigator had prior knowledge of the adherence literature was arguably the main source of bias. This is perhaps reflected in the language used within the labelling of some of the core categories within the current study (e.g., regimen characteristics) which is representative of many of the previous research findings on adherence to treatment. It is possible that someone with less knowledge of the existing literature may have used more novel or interpretive language to describe these observed phenomena.

4.8.1.2 Recruitment bias

The detailed process of recruitment was previously outlined in section 2.5. Participants were recruited by the specialist CF physiotherapists on a first-come, first-served basis (*i.e., the patients who had their outpatient clinics scheduled at the earliest date were the first to be approached*). Although the NRES committee stipulated that recruitment should be carried out in this way by the physiotherapists, it resulted in the chief investigator being unable to oversee the recruitment process to ensure that the agreed protocols were being appropriately adhered to. It is therefore possible that recruitment biases may have occurred through participants being selected by the physiotherapists in a less random way. However, the chief investigator was in regular contact with the specialist CF physiotherapists throughout the process of recruitment and was assured that the agreed protocols were being followed. The fact that the clinicians involved with the study advised that adolescent participants with an FEV1 reading <50% over the last 12 months should be excluded may have resulted in additional biases in terms of only reasonably healthy CF patients taking part.

Further reflection on the access to potential participants and the impact this may have had on the sample is provided in section 4.8.1.5.

4.8.1.3 Additional quality checks

Details of the quality checks employed and the additional strategies considered by the chief investigator were provided in section 2.9.2. Although estimates of inter-rater reliability were considered as a method of quality control in the current study, it was discounted on the grounds that numerous commentators have suggested that the technique is flawed for use within qualitative studies (Bogdan & Biklen, 1992; Marshall & Rossman, 1995). However, respondent validation is considered a useful technique in providing additional quality assurance (Smith, 2008) and the fact that the current study was unable to utilise this technique due to pragmatic concerns, is a weakness.

4.8.1.4 Participants access to internal processes

One important issue for all qualitative methodologies that rely on narrative accounts is that the participants may not actually be aware of the numerous internal and external influences on their behaviour (Corbin & Strauss, 2008). The result is that commonly used data collection methods, such as interviews, can only ever access information that the participant is consciously aware of. Even then, these accounts are only ever based on the participants' *perceptions* (Charmaz, 2006). A related issue is that participants may be consciously aware of a number of influences on their behaviour, but be unwilling to share them with the researcher. This relates to the issues highlighted in section 4.8.1 in that additional bias may be created by the effect the researcher's mere presence has on the people that they are studying.

Although it was clear that the participants in the current study spoke openly and provided rich and detailed accounts, the above difficulties cannot be discounted. These issues may have been particularly present in the adolescent accounts in that their developmental stage arguably results in a more "surface level" understanding of complex phenomena like adherence behaviour. In addition, the researcher became aware both during the interview process and throughout analysis that although adolescent participants appeared relaxed and forthcoming, they may have been somewhat reluctant to discuss their parents' behaviour with a stranger.

4.8.1.5 Sample

There are a number of issues to consider in relation to the sample of participants used within any form of qualitative research. Firstly, to ensure that participants are discussing their experiences of similar phenomena from a similar perspective, some argue for maximum homogeneity within a participant sample (Willig, 2008). Although many of the participants within the current study shared similar characteristics (e.g., all diagnosed with CF or caring for someone with CF, all adolescent participants aged between 11 and 16, all live within the North of England), there were also observed differences within both the patient and parent group. For example, some of the adolescent participants had co-morbid conditions such as CF-related diabetes, arguably making their experience of adhering to treatment different to those without a co-morbid condition. Some of the parent participants worked full-time, some part-time and some did not work at all, arguably making their experience of daily time pressures different from each other. In addition, although it was expected that mothers would primarily take part in the research (Timko et al., 1992), one father took part. Again, his

experience of a caring for his child is likely to be different to the experiences of the mothers taking part in the study.

One obvious inherent weakness of a homogenous sample, however, is that the smaller the sample and the more tightly defined their target characteristics, the less possible it becomes to generalise the study results beyond that specific sample (Smith, 2008). Although this may be true, it is also arguably the case for most qualitative research where the small sample sizes and emphasis on detailed individual accounts (regardless of homogeneity) make generalisability difficult. The current study interviewed six adolescent patients and six parents, resulting in a total sample size of 12. Although this is considered acceptable for grounded theory research (whereby the emphasis is on a rich and detailed account of a limited number of participants), a smaller sample size results in only tentative recommendations (in terms of the applicability of the findings to clinical settings), being able to be made.

Another consideration is that although the recruitment of participants was largely random (see section 2.5), it may well be possible that only “keen” parents and adolescents took part in the study, resulting in experiences being discussed from very particular perspectives (e.g., those who were proud of their adherence efforts and wanted recognition for this or conversely, those who had real difficulties maintaining adherence and so were desperate to “try anything” to improve the situation). Although this is always difficult to guard against or even to ascertain, it cannot be discounted as being a potential source of bias within the current study.

4.8.2 Methodological strengths

4.8.2.1 A detailed focus

The design of the current study enabled a detailed, more focussed exploration of one part of the CF treatment regimen. Much of the previous literature has examined adherence to the CF treatment regimen more generally (George et al., 2010). Although there are studies that have investigated adherence to specific components (e.g., Williams et al., 2007), adherence to aerosol therapy, particularly within the adolescent CF population has not been widely researched. This is problematic given the prominence of aerosolised treatments in current CF drug-delivery regimens and the likely increase of their use in the future. It is therefore considered a strength that the current study enabled a detailed exploration of the experiences of adhering to aerosol therapy.

4.8.2.2 Patient and parent perspectives

As well as providing a more detailed focus, the current study enabled experiences of adhering to aerosol therapy to be considered from two different perspectives. The patient experience is an obvious imperative, but the important influence of the parent on adherence to treatment has also been widely documented (Foster et al., 2001; Modi et al., 2008). Although adolescent adherence to the CF treatment regimen is often poor (Zindani et al., 2006; Riekert, et al., 2007), less is known about the potential influence of parents on the process of adhering to aerosol therapy during this key developmental stage. It is considered a strength that the current study enabled both a patient and a parent perspective concerning the process of adhering to aerosol therapy to be gained.

4.8.2.3 The use of the PLS data

The ability of the I-neb™ device to provide an objective and accurate measure of adherence to aerosolised therapy treatments enabled accounts of adherence to be anchored in everyday life and understood in the context of day-to-day behaviour. This was not only novel, but enabled adherence behaviour to be explored *in situ*, as close as possible to its actual occurrence, and thus, to an extent, reduce retrospective biases (Reis & Gable, 2000). The objectiveness of the data may have also helped to reduce subjective biases and potential social desirability effects (i.e., the tendency of participants to answer questions in a manner that will be viewed favourably by others).

A common criticism of qualitative methodologies is that they can only ever access information that the participant is consciously aware of. The fact that the PLS data provided a cue to participants about the actual rates of adherence, and prompted discussion of particular events helped to bring into conscious awareness the multiple influences on adherence.

4.8.2.4 The Grounded Theory Methodology

The use of the GTM enabled a systematic approach to the gathering and analysis of the generated data. The use of this methodical approach alongside regular research supervision enabled the maintenance of an audit trail which helped to ensure methodological and procedural rigour and reduce potential bias. As such, the resulting theoretical formulation was firmly grounded in the data. The ability of the GTM in developing a diagrammatic, theoretical formulation which helped to explain the multiple and interacting processes involved in adhering to aerosol therapy is itself considered a strength of the current

study. It is hoped that by providing a verbal narrative of the theoretical formulation, the chief investigators understanding is represented in a way that “achieves coherence and integration while preserving nuances in the data” (Elliott, Fischer, & Rennie, 1999).

The GTM encourages the researcher to “own one’s perspective” by recognising and disclosing their interests, values and assumptions and the role that these play in influencing understanding (Strauss & Corbin, 1990). By doing this (see section 2.9.3.1), it is hoped that the reader of the current study is able to interpret the data and the researchers’ understanding of them, and to consider possible alternatives. The GTM also encourages a description of the research participants and their life circumstances. In providing detailed pen portraits of the participants in the current study, it is hoped that the reader will be assisted in judging the range of persons and situations to which the findings might be relevant.

4.9 Clinical implications

The results of the current study have been considered in relation to the existing research literature. Although the smaller sample size means that the findings can only be used to make tentative recommendations for clinical practice, the following section will briefly highlight some of the main clinical implications of the research.

4.9.1 The aerosol therapy treatment regimen

Health care professionals should demonstrate a continued awareness of the burden associated with aerosol therapy as only one part of an already complex, demanding and often interdependent treatment regimen. Health professional efforts to ensure that less necessary additional treatments are kept to a minimum appear to be valued highly by patients and parents. Where increasing the number of aerosolised therapies is an absolute necessity, providing appropriate empathy and exploring and advising how the new regimen may fit into their lives may help to motivate patients and parents, particularly where the beliefs in the benefits associated with the aerosol therapy are less strong.

The utility of an M.I. approach (Miller, 1983; Miller & Rollnick, 1981) in facilitating these and other patient-health professional and parent-health professional interactions could prove worthwhile. It may be appropriate for training in this well researched approach to be encouraged for CF staff teams, as is already the case in some areas (Chiesi Connect, 2011).

4.9.2 *The aerosol therapy treatment device*

Advances in AAD technology have resulted in intelligent devices that are more effective, easier to use and better able to provide useful feedback that can both encourage adherence (e.g., reduced time associated with the TIM mouthpiece) and promote appropriate administration technique (e.g., cascading feedback system). Additional benefits are found in the ability of some of these newer devices (e.g., the I-neb™) to provide detailed and more objective adherence data than ever before. The current study extends the findings of those previously conducted (Latchford et al., 2009; McNamara et al., 2009) by suggesting that as well as providing an objective measurement of adherence, this information can be successful used to assist in the empathic and detailed exploration of potential barriers and facilitators of adherence to aerosol therapy. Gaining this in-depth understanding may be important for clinicians in helping patients and their parents to identify the numerous interacting biological, psychological and social influences on adherence to aerosol therapy, many of which were reported within the current study. In doing so, advice and support may be more appropriately targeted.

However, the benefits of more advanced devices paradoxically create a number of associated difficulties. Ensuring that the device is appropriately maintained and cleaned creates an additional burden which was identified as a potential barrier by participants in the current study. The negative impact that this had on device portability and associated non-adherence was a particularly relevant concern. In addition, some of the participants' perceptions of the I-neb™ as fragile and expensive (and the associated fear of losing or damaging the device) further prevented the aerosol therapy from being adhered to outside of the home environment.

It should perhaps be encouraged for appropriate stakeholders (i.e., patients, parents, clinicians and device manufacturers) to engage in a *cost-benefit analysis*, that enables an open account of the benefits of continued technological device improvements alongside the associated consequences. In doing so, the rhetoric of continual advancement can be more appropriately situated in the context of everyday life.

4.9.3 *Cognitive, affective and behavioural processes*

The current study found that a wide variety of patient and parent beliefs in the aerosol therapy had the potential to influence a series of adaptive and less adaptive affective and behavioural responses. The impact of patient cognitions and beliefs on the process of adherence is well documented and was reviewed earlier (e.g., Horne, 2003). Assisting

patients and parents in the identification of these interacting psychological processes (perhaps utilising the PLS data where deemed appropriate) may help to target appropriate support, particularly for those whose problematic cognitions (e.g., a parent thinking that the aerosol therapy is not working) and emotions (e.g., a parent feeling anxiety and guilt) have the potential to result in less helpful behaviours (e.g, excessively reminding or “nagging” their child to take the aerosol therapy) that may negatively impact on the parent and child relationship.

M.I. is one potential approach to assist clinicians in this task, as are cognitive behavioural therapy (CBT) approaches, which are now beginning to demonstrate their effectiveness within the wider adherence literature (Duff & Latchford, 2010a). Again, it may be appropriate for training in these approaches to be encouraged.

4.9.4 The parent and adolescent relationship

The current study highlighted the importance of a positive parent-adolescent relationship both generally and in its ability to act as a “buffer” to counteract some of the daily struggles associated with adhering to aerosol therapy. This echoes the findings of countless previous research studies, many of which were reviewed earlier. The current study findings suggest that particular concerns may centre around the transfer of adherence responsibility, either prematurely (i.e., giving the adolescent too much responsibility for managing the process of adhering to aerosol therapy too soon) or inappropriately delayed (i.e., failing to provide adolescents with the type of supported autonomy that would be appropriate for their developmental stage).

Clinician’s may feel less able to affect change for those patients and parents where less positive aspects of their relationship are negatively impacting on the process of adherence. However, an empathic and understanding approach may allow patients and parents to feel able to discuss these difficulties more openly.

4.9.5 The context of adherence

The current study emphasised that adherence to aerosol therapy does not exist in a vacuum. The use of the PLS data within the semi-structured interviews enabled a number of lifestyle and environmental influences to be identified. Again, while clinicians may feel less able to intervene to bring about change in any problematic areas, the open discussion of these influences (once more, using the PLS data where deemed appropriate) may help to identify

specific commitments, times, events or situations which have the potential to negatively impact on the process of adhering to aerosol therapy.

One overwhelming finding in the current study was that the interacting biopsychosocial processes described above make the prospect of consistent, one hundred percent adherence to aerosol therapy very unlikely. Clinicians may need to openly assess their own reactions to this. Although clinicians are a valuable source of support and may be able to help affect positive change, it would appear that non-adherence to a treatment they know can help to prolong both the quality and longevity of their patients' lives is perhaps an unfortunate inevitability.

4.10 Implications for future research

4.10.1 The strengths and limitations of nebuliser technology

Advances in AAD technology have resulted in more intelligent devices. However, the current study found that the very real benefits of more advanced devices paradoxically create a number of associated difficulties. The cleaning of the device, for example was identified as a particular concern. Studies that examine both the strengths and limitations of nebuliser devices would enhance understanding and may assist in further product development.

4.10.2 The use of M.I.

The current study highlighted a number of difficulties (e.g., the multiple biopsychosocial interacting influences on adherence to aerosol therapy) that could potentially be explored with adolescents and parents during routine CF clinic visits. The approach taken by clinicians is vitally important in determining how successful or otherwise these interactions might be (Becker & Maiman 1975; Squier, 1990). Although M.I. approaches have been well documented within the literature (Duff & Latchford, 2010a) further research examining M.I. for CF adherence difficulties would help to ascertain their utility within routine clinical practice.

4.10.3 The use of diary studies

The current study demonstrated the usefulness of the PLS data downloaded from the I-neb™ in helping to qualitatively explore the process of adhering to aerosol therapy. The PLS data was used as a type of diary which helped to cue reflections on actual events around

adherence. The use of diary methodologies (i.e., sequentially collecting data on or from individuals within a temporal framework) is becoming commonplace in clinical and health psychology research (Ferguson, 2005). While this method has been previously used to examine more general adherence behaviour in CF (e.g., Modi, & Quittner, 2006), studies which utilise diary data to examine specific components of the treatment regimen (e.g., aerosol therapy) may help to further enhance understanding of the likely multiple, daily barriers and facilitators to adherence.

4.10.4 Psychodynamic and systemic approaches

The continued emphasis on research studies in examining largely cognitive, conscious processes such as “illness cognitions” and “health beliefs” has contributed to the paucity of research from alternative theoretical perspectives. The current study found that adherence behaviours such as “forgetting” for adolescents or “reminding” for parents may have a more unconscious foundation than is commonly reported within the literature. Although there are examples of studies examining more psychodynamic approaches to adherence (e.g., Alfonso, 2011), further research in this area may enhance understanding and help to shift the focus away from a largely cognitive approach.

Similarly, the current study highlighted the numerous social, structural and environmental influences on adherence. Although these findings are not particularly surprising, some have argued that research from more systemic approaches would enable a better understanding of the way in which social and environmental factors influence adherence (Frohlich et al., 2001).

5.0 CONCLUSION

In response to the research questions and aim, the following conclusions can be drawn:

1. Although the process of adhering to aerosol therapy is experienced very differently by the parents and adolescents, their narrative accounts were actually very complementary. This was reflected in the theoretical formulation which was so similar because participants talked about the same material but often from very different perspectives.
2. Although the results identified much that was expected, the theoretical formulation adds a richer account and shows how these different biopsychosocial processes potentially interact and influence adherence to aerosol therapy.
3. There were also more novel findings including:
 - the way that certain treatments are interrelated (e.g., aerosol therapy and physiotherapy) and the associated burden that this creates which can negatively impact on the process of adherence
 - the way that technological advances in the manufacturing of nebuliser devices paradoxically create associated difficulties (e.g., the necessity of cleaning the device; the device perceived as fragile)
 - the way in which participants are influenced by an internal hierarchy of preference for certain CF treatments and the interesting and sometimes surprising reasons for this (e.g., preferring certain types of physiotherapy because friends can get involved; disliking aerosol therapy because it isolates them from their friendship group)
 - the belief, particularly from the parents, that non-adherence to aerosol therapy is an unfortunate inevitability because of the influence of the reported interacting biopsychosocial processes
 - the seemingly impossible task of achieving an appropriate balance (e.g., adolescents adhere more to their aerosol therapy when they are reminded by their parents, but also resent this)
4. The PLS data downloaded from the I-neb™ device provided a cue to participants about the actual rates of adherence, and prompted discussion of particular events. This helped to bring into conscious awareness the multiple interacting influences on adherence to aerosol therapy.

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APPENDICES

Appendix 1 – Letter of ethical approval



National Research Ethics Service

NRES Committee Yorkshire & The Humber - Humber Bridge

Yorkshire and the Humber Research Ethics Office
First Floor
Millside
Mill Pond Lane
Leeds
LS6 4RA

Telephone: 0113 3050127

12 September 2011

Mr Daniel P H O'Toole
Psychologist in Clinical Training
The Leeds Teaching Hospitals NHS Trust
Leeds Institute of Health Sciences
Charles Thackrah Building
101 Clarendon Road, Leeds
LS2 9LJ

Dear Mr O'Toole

Study title: Experiences of adhering to aerosol therapy in adolescents with cystic fibrosis: patient and parent perspectives.

REC reference: 11/YH/0284

Thank you for your letter of 02 September 2011, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Vice-Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS sites

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

This Research Ethics Committee is an advisory committee to the Yorkshire and The Humber Strategic Health Authority
The National Research Ethics Service (NRES) represents the NRES Directorate within
the National Patient Safety Agency and Research Ethics Committees in England

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rdforum.nhs.uk>.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Covering Letter		27 June 2011
Evidence of insurance or indemnity		
GP/Consultant Information Sheets		15 August 2011
Interview Schedules/Topic Guides		24 June 2011
Interview Schedules/Topic Guides	1	24 June 2011
Investigator CV		24 June 2011
Other: CV - G Latchford (supervisor)		04 July 2011
Participant Consent Form: Consent to be contacted form for parent/guardian	1	20 June 2011
Participant Consent Form	2	15 August 2011
Participant Information Sheet: Parent/Guardian	2	15 August 2011
Participant Information Sheet: Young Person 11 - 13	2	15 August 2011
Participant Information Sheet: Young Person 14 - 16	2	15 August 2011
Protocol	1	24 June 2011
REC application		04 July 2011
Response to Request for Further Information		02 September 2011

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "*After ethical review – guidance for researchers*" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

Further information is available at National Research Ethics Service website > After Review

11/YH/0284

Please quote this number on all correspondence

With the Committee's best wishes for the success of this project

Yours sincerely



Dr **Dr David Horton**
Chair

Email: nicola.mallender-ward@nhs.net

Enclosures: "After ethical review – guidance for researchers"

Copy to: Rachel E de Souza, University of Leeds
Dr Derek Norfolk, Leeds Teaching Hospitals NHS Trust

Appendix 2 – Young person's information sheet about the research

**Leeds Institute of
Health Sciences**

FACULTY OF MEDICINE AND HEALTH



UNIVERSITY OF LEEDS

Young person's information sheet about the research (11 to 13 Years) **Version 2 (15.08.11)**

Title of Research Study: Experiences of adhering to aerosol therapy in adolescents with cystic fibrosis: patient and parent perspectives.

You are invited to take part in a research study conducted by Danny O'Toole (Psychologist in Clinical Training) as part of my doctorate degree at the University of Leeds. Before you decide whether or not you would like to take part in this research, please read the following information carefully about why the research is being done and what it would involve for you. You can take as much time as you need to decide. You can also talk to other people about it like your family, friends or staff at the hospital if you want to.

What is the purpose of the study?

There have been hardly any studies which have looked at what it is like to take aerosol therapy for someone with cystic fibrosis (CF). Some people take their aerosol therapy regularly as prescribed by their doctors while others find it more difficult. I am interested in finding out about your experiences of taking aerosol therapy and the experiences of your parent/guardian in supporting you to take your aerosol therapy.

Why have we been invited to take part?

You have been chosen to take part because you have CF, you have been using your aerosol therapy device for at least the last 12 months and you are aged between 11 and 16. I would also like to talk to one of your parents or the person who looks after you about their experiences.

Do we have to take part?

You do not have to take part in this study; it is up to you to decide. If you do decide to take part you will be asked to sign a form to show that you agree to take part. Your parent or guardian will also need to sign this form to show that they agree to take part.

You can stop taking part in the study at any time if you change your mind, without having to say why. If you decide not to take part or to stop part way through, this will have no effect on the care you get from the team at the hospital.

What will happen to us if we take part?

If you decide you want to take part, I will telephone you to answer any questions you may have and to arrange a time to come and meet both you and your mum, dad or the person who looks after you at your home. I will also arrange for 1 weeks worth of the information that is regularly downloaded from your aerosol therapy device at your next outpatient's appointment to be sent to me ahead of our meeting so that we can use it when we speak to each other to help to jog your memory. This information will be sent to me safely by e-mail and will not have your name or any personal details about you contained within it. Before this is sent to me, both you and your mum, dad or the person who looks after you will need to sign a form which says that you agree for this information to be sent.

When we meet at your home, I will interview both you and your mum, dad or the person who looks after you separate from each other. I will ask you questions about what it is like taking your aerosol therapy and will ask your mum, dad or the person who looks after you questions about what it is like trying to support a young person who has to take aerosol therapy. Each interview should last about 60 minutes, depending on how much you have to say.

Although we will try to do the interviews separately, you may want someone else to sit in with you for support. This might be another adult member of your family or even a friend. Remember you can also stop the interview and withdraw from the study at any time, without giving a reason.

I will tape record both conversations. This is because afterwards, everything you have said on the tape will be written down. This will allow me to look at what you have both said, and try to find the main issues that you talked about.

After I have looked at all the interviews, I will write to everyone who has taken part in the research with a summary of what I thought the main issues were.

What are the possible advantages or disadvantages of taking part?

If you take part I cannot promise that this study will have any direct benefit for you. However, some people may find that talking to someone about their experiences helpful. If you do take part, you and your mum, dad or the person who looks after you will receive a £15 gift voucher to share.

Although talking about your experiences of taking aerosol therapy is unlikely to cause you to become upset, if you do find that you become upset during the interview I can let your parent know, if you say that is okay. If you would like, I can also arrange for you to see a member of the CF care team at the hospital so that you will be able to talk to them about how you are feeling.

Will our taking part in the study be kept confidential?

I will ask you and your mum, dad or the person who looks after you to sign a form which says you agree to me writing to your G.P. (family doctor). This is just to let them know you are taking part in the study.

After the interview, everything you have said on the tape will be written down. Any information that might show who you are will be kept separately from this written

version, and they will both be kept in a safe place, until they are destroyed in 3 years time. The information that is downloaded from your aerosol therapy device will not have your name or any personal details about you included within it. This information will also be kept in a secure place, until they are destroyed in 3 years time.

I will not tell anyone else what you have said unless I am worried about you or someone else getting hurt in some way. If I am worried about this happening I will have to tell someone.

What will happen to the results of this study?

I will write up the results of this study for my studies. I will include some of the exact things people have said. It is also possible that I will present the results at a conference or write them down in a journal article. At all times, your privacy will be protected. I will do this by calling you a different name, which you can choose yourself if you want.

When I write up this research I will try my best to protect your privacy but because you and your parent/guardian know each other so well, you might think that you recognise each other from what is being said in the report. However, it is very unlikely that anyone else reading it will be able to tell who you are.

Who has reviewed this study?

Before any research goes ahead it has to be checked by an independent group of people called a Research Ethics Committee (REC). They make sure the research is fair. This study has been checked by the Humber Bridge REC. It has also been checked by staff at the University of Leeds.

Where can I find out more information?

If you would like more information, or have any questions about the study you can contact me (Danny O'Toole) or Dr Gary Latchford (my research supervisor) on the contact details below:

Danny O'Toole

Psychologist in Clinical Training
Clinical Psychology Training Programme
Leeds Institute of Health Sciences
Charles Thackrah Building
University of Leeds
101 Clarendon Road
Leeds
LS2 9LJ

Tel: 0113 343 0815

E-mail: undpho@leeds.ac.uk

Dr Gary Latchford

Consultant Clinical Psychologist
Clinical Psychology Training Programme
Leeds Institute of Health Sciences
Charles Thackrah Building
University of Leeds
101 Clarendon Road
Leeds
LS2 9LJ

Tel: 0113 343 2736

E-mail: g.latchford@leeds.ac.uk

If you would like to seek independent advice regarding taking part in this study, you can contact the Leeds Teaching Hospitals PALS (Patient Advice and Liaison Service) on 0113 206 7168)

Appendix 3 – Parent information sheet about the research

**Leeds Institute of
Health Sciences**

FACULTY OF MEDICINE AND HEALTH



UNIVERSITY OF LEEDS

Parent/guardian's information sheet about the research **Version 2 (15.08.11)**

Title of Research Study: Experiences of adhering to aerosol therapy in adolescents with cystic fibrosis: patient and parent perspectives.

You are invited to take part in a research study conducted by Danny O'Toole (Psychologist in Clinical Training) as part of my doctorate degree at the University of Leeds. Before you decide whether or not you would like to take part in this research, please read the following information carefully about why the research is being done and what it would involve for you. You can take as much time as you need to decide and you can talk to others about it if you wish.

What is the purpose of the study?

There have been hardly any studies which have looked at what it is like to take aerosol therapy for someone with cystic fibrosis (CF). Some people take their aerosol therapy regularly as prescribed by their doctors while others find it more difficult. I am interested in finding out about your child's experiences of taking aerosol therapy and your experiences of supporting your child to take aerosol therapy.

Why have we been invited to take part?

Your child has been chosen to take part because they have CF, they have been using their aerosol therapy device for at least the last 12 months and they are aged between 11 and 16. I would also like to talk to you about your experiences of supporting your child to take aerosol therapy.

Do we have to take part?

You do not have to take part in this study; it is up to you to decide. If you do decide to take part you will be asked to sign a consent form to show that you agree to take part and that you agree for your child to take part.

Both you and your child are free to withdraw from the study at any time, without giving a reason. Not taking part or withdrawing will have no effect on the standard of care you or your child receive from the team at the hospital.

What will happen to us if we take part?

If you decide you want to take part, I will telephone you to answer any questions you may have and to arrange a time to come and meet both you and your child to carry out an interview at your home. I will also arrange for 1 weeks worth of the information that is routinely downloaded from your child's aerosol therapy device at

your next outpatient's appointment to be sent to me ahead of our meeting so that we can use it in the interview to help to jog yours and your child's memory. This information will be sent to me via secure e-mail correspondence and will not have any personally identifiable details about you or your child contained within it. Before this is sent to me you will need to sign a consent form to show that you agree to this being done.

When we meet at your home, I will interview both you and your child separate from each other. I will ask your child questions about what it is like taking their aerosol therapy and will ask you questions about what it is like trying to support your child in taking their aerosol therapy. Each interview should last about 60 minutes, depending on how much you have to say.

Although we will aim to do the interviews separately, it is fine for someone else to sit in with your child for support. This might be another adult member of your family or even a friend, depending on who they prefer. Remember that you or your child can stop the interview and withdraw from the study at any time, without giving a reason.

I will tape record both conversations. This is because afterwards, everything you have said on the tape will be transcribed. This will allow me to look at what you and your child have said, and try to find the main issues that you raised.

After I have analysed all of the interviews, I will write to everyone who has taken part in the research with a summary of what I thought the main issues were.

What are the possible advantages or disadvantages of taking part?

If you decide to take part I cannot promise that this study will have any direct benefit for you or your child. However, some people may find that talking to someone about their experiences helpful. If you do take part, you and your child will receive a £15 gift voucher to share.

Although talking about experiences of aerosol therapy is unlikely to cause you or your child distress, if you or your child do find that you become upset during the interview I can arrange (with your permission) for either or both of you to see a member of the CF care team at the hospital so that you will be able to talk to them about how you are feeling. They may be able to offer you further support and advice.

Will our taking part in the study be kept confidential?

I will ask you to sign a form which says that you agree to me writing to your child's G.P. This is just to let them know you are taking part in the study.

After the interview, everything you have said on the tape will be transcribed. Any information that identifies who you are will be kept separately from these interview transcripts and both sets of information will be kept in a secure location, until they are destroyed in 3 years time. The information that is downloaded from your aerosol therapy device will not have any personally identifiable details about you or your child contained within it. This information will also be kept in a secure place, until they are destroyed in 3 years time.

I will not tell anyone else what you or your child says within the interview unless I am concerned about the safety of you, your child or anyone else. If I am concerned, I will have to talk to my supervisors and seek further advice.

What will happen to the results of this study?

I will write up the results of this study for my doctorate. I will include some direct quotes of what people have said. It is also possible that I will present the results at a conference or publish them in a journal article. At all times I will protect your anonymity and that of your child by using pseudonyms (i.e., I shall refer to you both by names other than your own). You and your child may choose these alternative names if you wish.

When I write up this research I will make every effort to protect your anonymity by withholding identifying information and using pseudonyms but because you and your child know each other so well, you might think that you recognise each other from what is being quoted should you read any of my reports. However, it is very unlikely that anyone else reading the work will be able to identify either of you.

Who has reviewed this study?

All research in the NHS has to be assessed by an independent Research Ethics Committee (REC) to ensure your rights, dignity and well-being is protected. This study has been assessed by the Humber Bridge REC. It has also been reviewed by two academic panels at the University of Leeds.

Where can I find out more information?

If you would like more information, or have any questions about the study you can contact me (Danny O'Toole) or Dr Gary Latchford (my research supervisor) on the contact details below:

Danny O'Toole

Psychologist in Clinical Training
Clinical Psychology Training Programme
Leeds Institute of Health Sciences
Charles Thackrah Building
University of Leeds
101 Clarendon Road
Leeds
LS2 9LJ

Tel: 0113 343 0815

E-mail: undpho@leeds.ac.uk

Dr Gary Latchford

Consultant Clinical Psychologist
Clinical Psychology Training Programme
Leeds Institute of Health Sciences
Charles Thackrah Building
University of Leeds
101 Clarendon Road
Leeds
LS2 9LJ

Tel: 0113 343 2736

E-mail: g.latchford@leeds.ac.uk

If you would like to seek independent advice regarding taking part in this study, you can contact the Leeds Teaching Hospitals PALS (Patient Advice and Liaison Service) on 0113 206 7168)

Appendix 4 – Consent form

Consent form for Parent/Guardian – Version 2 (15.08.11)

Title of Research Study: Experiences of adhering to aerosol therapy

Name of Researcher: Danny O'Toole

Please initial the
boxes if you agree

- | | | |
|----|---|--------------------------|
| 1. | I confirm that I have read and understand the information sheet dated 15.08.11 (version 2) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. | <input type="checkbox"/> |
| 2. | I understand that my participation and that of my child is entirely voluntary. I understand that we are entitled to withdraw from the study at any time without giving any reason and without our medical care or legal rights being affected. | <input type="checkbox"/> |
| 3. | I agree to the last 7 days of adherence data which has been downloaded as routine at this out-patient appointment being sent to the above named researcher via secure e-mail correspondence. I understand that this data will not contain any personally identifiable information within it. I understand that this data will only be used for the purpose of the interview and nothing else. | <input type="checkbox"/> |
| 4. | I give permission for the interview to be audio-recorded. | <input type="checkbox"/> |
| 5. | I give permission for a letter to be sent to my child's G.P. advising of our involvement in the above study. | <input type="checkbox"/> |
| 6. | I understand that any information I offer will be treated anonymously and all material arising out of the study will be dealt with on a confidential basis by the researcher involved. The research complies with the Data Protection Act (1998). | <input type="checkbox"/> |
| 7. | I understand that data collected during the study may be looked at by individuals from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to this information. | <input type="checkbox"/> |
| 8. | I agree to participate in the above named study. | <input type="checkbox"/> |
| 9. | I agree to my child taking part in the above named study. | <input type="checkbox"/> |

Name of child
participating

Date

Signature

Name of parent/
guardian participating

Date

Signature

Name of health
Professional taking
consent

Date

Signature

Appendix 5 – Adolescent patient interview schedule

Semi-structured Interview Guide – Child **Version 1 – 24.06.11**

Aim: Approximately 8 – 10 key questions per hour session

Q's Should be: Direct, Forthright, Comfortable, Simple, Clear, Brief, Reasonable, Jargon-free, Short (i.e. one-dimensional). Exact wording may change as interviews progress.

Question type:

- Initial open-ended
- Intermediate
- Ending

(Charmaz, 2006)

Interview Question	Prompts & Topic Guide	Question Type
Pen portrait and introduction questions?	<ul style="list-style-type: none"> • How old are you? • What school do you go to? 	Initial Open-ended
Describe a typical day for you?	<ul style="list-style-type: none"> • What types of things do you do? 	Initial Open-ended
Tell me a bit more about what it is like living with CF?	<ul style="list-style-type: none"> • “Think back” • Good/bad • Impact • Family Relationships • Friends • Treatments 	Intermediate
Can you tell me a bit more about the treatments you take for CF?	<ul style="list-style-type: none"> • Which ones? • What are they for? • What are they like? • Which do you prefer? 	Intermediate
What is it like having to take your aerosol therapy?	<ul style="list-style-type: none"> • Like/dislike? • Easy/hard? 	Intermediate
What things make it easier for you to take your aerosol therapy?	<ul style="list-style-type: none"> • Think back to a particular time... • Help/support • Being reminded • Having time • Any days/times when it's easier 	Intermediate
Looking at this information here, were there any days and times that you found it easier to take your aerosol therapy?	<ul style="list-style-type: none"> • Use the I-neb data to prompt • Tell me a little about day X • What was it about day X/time Y that helped you 	Intermediate

What things make it difficult for you to take your aerosol therapy?	<ul style="list-style-type: none"> • Think back to a particular time... • Being really busy • Forgetfulness • Not liking it 	Intermediate
Looking at this information here, were there any days and times that you found it more difficult to take your aerosol therapy?	<ul style="list-style-type: none"> • Use the I-neb data to prompt • Tell me a little about day X • What was it about day X/time Y that was difficult 	Intermediate
What advice would you give to someone who is finding it hard to take their aerosol therapy?		Ending
You've told me that (summarise). Is there anything else you think I should know to help me understand more about what it is like having to take aerosol therapy?		Ending

Thank you for your time

Remember to record impressions after the interview

Appendix 6 – Parent interview schedule

Semi-structured Interview Guide - Parent **Version 1 – 24.06.11**

Aim: Approximately 8 – 10 Questions per hour session

Q's Should be: Direct, Forthright, Comfortable, Simple, Clear, Brief, Reasonable, Jargon-free, Short (i.e. one-dimensional). Exact wording may change as interviews progress.

Question type:

- Initial open-ended
- Intermediate
- Ending

(Charmaz, 2006)

Interview Question	Prompts & Topic Guide	Question Type
Pen portrait and introduction questions?	<ul style="list-style-type: none"> • How old are you? • What do you do for a job? 	Initial Open-ended
Describe a typical day for you?	<ul style="list-style-type: none"> • What types of things do you do? 	Initial Open-ended
Tell me a bit more about what it is like having a child with CF?	<ul style="list-style-type: none"> • “Think back” • Good/bad • Impact • Family Relationships • Friends • Treatments 	Intermediate
Can you tell me a bit more about the treatments your child takes for CF?	<ul style="list-style-type: none"> • Which ones? • What are they for? • What are they like? • Which do they prefer? • Which do you prefer? 	Intermediate
Tell me some more about your child's aerosol therapy?	<ul style="list-style-type: none"> • Like/dislike? • Easy/hard? 	Intermediate
What things make it easier for your child to take their aerosol therapy?	<ul style="list-style-type: none"> • Think back to a particular time... • Help/support • Being reminded • Having time • Any days/times when it's easier 	Intermediate
Looking at this information here, were there any days and times that you think your child found it easier to take their aerosol therapy?	<ul style="list-style-type: none"> • Use the I-neb data to prompt • Tell me a little about day X • What was it about day X/time Y that helped your child 	Intermediate

	<ul style="list-style-type: none"> • Is that a common pattern 	
What things make it difficult for your child to take their aerosol therapy?	<ul style="list-style-type: none"> • Think back to a particular time... • Being really busy • Forgetfulness • Not liking it 	Intermediate
Looking at this information here, were there any days and times that you think your child found it more difficult to take their aerosol therapy?	<ul style="list-style-type: none"> • Use the I-neb data to prompt • Tell me a little about day X • What was it about day X/time Y that was difficult for your child • Is that a common pattern 	Intermediate
What advice would you give to a parent whose child is finding it hard to take their aerosol therapy?		Ending
You've told me that (summarise). Is there anything else you think I should know to help me understand more about having a child who takes aerosol therapy?		Ending

Thank you for your time

Remember to record impressions after the interview

Appendix 7 – Example of anonymised PLS diary data sheet

Date	Dose Prescribed	Dose Taken	Drug Dose	Time Taken	Duration
Wednesday 1 st February	2	1	Full	08:08 (8.08am)	4 mins
		1	Full	22:14 (10.14pm)	5 mins
Thursday 2 nd February	2	1	Full	18:39 (6.39pm)	4 mins
		1	Full	21:00 (9.00pm)	4 mins
Friday 3 rd February	2	1	Full	08:04 (8.04am)	4 mins
		1	Full	21:47 (9.47pm)	4 mins
Saturday 4 th February	2	1	Full	10:59 (10.59am)	6 mins
		N/A	N/A	N/A	N/A
Sunday 5 th February	2	1	Full	13:47 (1.47pm)	4 mins
		1	Full	20:18 (8.18pm)	4 mins
Monday 6 th February	2	1	Full	07:53 (7.53am)	6 mins
		1	Full	21:06 (9.06pm)	4 mins
Tuesday 7 th February	2	1	Full	08:49 (8.49am)	3 mins
		1	Full	20:45 (8.45pm)	3 mins

Appendix 8 – Example of the open coding process

CHOOSING JOB INFLUENCED BY OF (1)
 DAYS HAVING DIFFERENT ROUTINES (2)
 DAD ARE BUSY (3)
 CHANGING ROUTINE (3)
 INES BEHAVING (4)
 HAVING WEB REARRANGED (4)
 INES IS EXPANSIVE (4)
 WEB IS FANTASTIC (4)
 INES MAKING A NOTICEABLE DIFFERENCE TO CHILD'S BEHAVIOUR (4)
 INES PREVENTING HOSPITAL ADMISSIONS (4)
 BEING TAKEN OFF WEB WAS FEARFUL (4)
 BEING TAKEN OFF WEB RESULTED IN HOSPITAL ADMISSION (4)
 BEING PUT BACK ON THE WEB (5)
 INES PREVENTING HOSPITAL ADMISSIONS (5)

OF HAVING CONSEQUENCES (6)
 GETTING ON WITH IT (6) (9)
 LIVING WITH IT (6)
 BEING DIAGNOSED WITH DIABETES (6)
 BEING PUT ON INSULIN (7)
 HAVING TO MONITOR BLOOD GLUCOSE (7)
 CHANGING NATURE OF TREATMENT REGIMEN (8)
 HAVING TO TAKE MULTIPLE TREATMENTS (8)
 HAVING TO DO TWICE A DAY (8)
 INES IS NOT IN ISOLATION TO OTHER TREATMENTS (8)
 BEING MORE EMBARRASSED IN A MORNING (9)
 BELIEVING PAIN IS IMPORTANT (9)
 HAVING FAITH IN HOSPITAL TEAM (9)
 BEING IN A ROUTINE (9)

HAVING TO IMPROVE (10)
 STRATEGIES FOR TREATMENT ARE VARIOUS ON HOW TO (10)
 FREQUENT TAKING RESPONSIBILITY FOR TREATMENT PROBLEMS (10)
 CHILD HAVING TO TAKE RESPONSIBILITY TO REMEMBER (10)
 BELIEVING CHILD TO CHILD (10)
 TRUSTING THE CHILD (10)
 TAKING WEB ON SUMMER CAMP (10)
 NEEDING A UTILE TO CLEAN WEB WITHOUT BURN (10)
 CLEANING WEB IS NECESSARY AFTER USE (10)
 CAMPING CREATES PROBLEMS FOR WEB ADHERENCE (10)
 NOT HAVING CLEANING FACILITIES WHILE CAMPING (10)
 CHILD ORIGINAL BEING PRESCRIBED REGIMEN (10)
 INES MAKING CHILD OFFER (11)
 GIVES PREVENTING PSEUDOMONAS (11)
 A.T. HELPING KEEP CHILD HEALTHY (11)

FITTING EVERYTHING IN (12)
 HAVING A ROUTINE (13)
 ADJUSTING THE ROUTINE TO SUIT (13)
 FORGETTING TO TAKE WEB (13)
 FORGETTING HAS CONSEQUENCES (13)
 ESTABLISHING ROUTINES (13)
 SPLITTING THE RESPONSIBILITY (13)
 PARENTS COOPERATING (13)
 HAVING A CONVERSATION ABOUT ROUTINE (13)
 BEING IN BEDTIME (13)
 FITTING WEB AROUND LIFESTYLE (14)
 PREPARING A ROUTINE (14)
 ATTENDING TO A ROUTINE (14)

BEING IN A ROUTINE (5)
 CHILD NEEDING TO BE PROMPTED (5)
 CHILD NEEDING TO BE REMINDED TO TAKE WEB (5)
 REMINDING IS SOMETIMES NECESSARY (5)
 DEVELOPING CHILD (5)
 CHILD TAKING MORE CONTROL (5)
 CHILD PREPARING AND OWN TABLETS (5)
 PLANNING + PREPARING TABLETS AT BEGINNING OF WEEK (5)
 GETTING FASTER AS CHILD GETTING OLDER (5)
 CHILD GETTING OLDER AT IT (5)
 CHILD HAVING GOOD BEHAVIOUR (5)
 CHILD TAKING TABLETS IN FRONT OF PARENTS (5)
 PARENTS ASKING CHILD QUESTIONS ABOUT IT (5)
 CHILD HAVING A CONSISTENT EXERCISE GROUP (5)

MAKING PAIN FUN (9)
 WATCHING TV WHILE GOING PAIN (9)
 TREATMENTS ARE INTERDEPENDENT (9)
 BEING AT HOME IS EASIER FOR TREATMENT (9)
 GOING ON HOLIDAY CREATES CONSEQUENCES (9)
 BEING WITHOUT SOME COMFORTS (9)
 NOT HAVING A SOLID VOTER (9)
 BEING ADVISED BY THE HOSPITAL RE: HOW ADHERENCE WITHOUT ON HOLIDAY (9)
 HOSPITAL BEING FLEXIBLE WITH ADH. ADHERENCE (9)
 EXPERIMENTING WITH ADH. ADHERENCE (9)
 BEING ABLE TO TOLERATE A PERIOD OF INES NOW ADHERENCE (9)
 INES IS PERSISTENT (9)
 BEING ABLE TO TAKE IT WITH US IF WE GO ON TRIP (9)

TAKING CHILD OFF WEB RESULTS IN HOSPITAL ADMISSIONS (11)
 BEING INTRODUCED TO THE TIM MOUTHPIECE (11)
 OLDER MOUTHPIECE TAKING LONGER (11)
 TIM MOUTHPIECE REDUCES TIME SIGNIFICANTLY (11)
 TAKING A.T. OUTCOMES (12)
 TIM MOUTHPIECE GIVES CHILD BACK TIME IN THE EVENING (12)
 DOING WEB LAST THING AT NIGHT (12)
 TREATMENTS ARE INTERDEPENDENT (12) (13)
 TREATMENTS ARE TIME CRITICAL (12) (13)
 NEEDING AN HOUR BETWEEN TREATMENTS (12)
 BELIEVING IN A.T. (12)
 CHANGING ROUTINE (12)
 BEING FLEXIBLE (12) (13)

STICKING TO A ROUTINE IS POSITIVE (14)
 CHAOTIC ROUTINES ARE DISORDERLY (14)
 BEING ON HOLIDAY IS DIFFICULT (14)
 NOT HAVING CLEANING FACILITIES ON HOLIDAY (14)
 BEING FLEXIBLE WITH ADHERENCE ON HOLIDAY (14)
 NEEDING A UTILE TO BOIL AND CLEAN WEB (14)
 NEEDING TO CLEAN WEB PROPERLY (14)
 BUILDING WEB INTO YOUR ROUTINE (14)
 SITTING DOWN AND DISCUSSING ADHERENCE AS A FAMILY (14)
 MAKING SURE ADHERENCE PLAN IS CONSISTENT (14)
 PROVIDING CHILD WITH A CONSISTENT ROUTINE (14)
 NEGOTIATING ADHERENCE BETWEEN PARENTS (14)
 FATHER TAKING RESPONSIBILITY FOR CHECKING (14)
 PARENTAL MONITORING + SUPERVISION (14)

[illegible]

Appendix 10 – Example of a diagram used during the parent data analysis

